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DOCTORAL THESIS

Lower Limb Alignment, Assessment and Management by Orthoses in Children with Cerebral Palsy.

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Lower Limb Alignment, Assessment and Management by Orthoses in Children with Cerebral Palsy

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Abstract

Cerebral Palsy (CP) is a neurodevelopmental condition well recognised to begin at birth or early childhood and persisting through the lifespan. The condition has been defined as a group of permanent disorders impacting on the development of movement and postures in turn causing activity limitation. Impacts caused by CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy and by secondary musculoskeletal problems.

Physiotherapists are allied health professionals who assist patients with optimising their health and wellbeing outcomes, such as musculoskeletal problems. These outcomes are achieved through evidenced-based care whereby physiotherapists assess, diagnose, and treat a wide range of health conditions throughout the lifespan. One discipline in physiotherapy pertains to the provision of care to the paediatric population, whereby the physiotherapist can assist in optimising a child's mobility and strength (for example, in children with CP), thereby reducing or even preventing other sequalae associated with their conditions (such as musculoskeletal discomfort and deformity). One treatment approach often take by physiotherapists is to prescribe lower limb orthoses, such as ankle-foot orthoses (AFOs). However, there is limited evidence on sensomotoric orthoses (SMotOs), which are used clinically in children with CP. As such, the research question underpinning this program of research was 'Which lower limb orthoses are optimal for treating children with CP?'. Therefore, the aims of this program of research were to: investigate current lower limb assessment techniques, determine what lower limb orthoses were available for children with CP, determine the effects of these lower limb orthoses on the gait and gross motor skills of children with CP, and to determine if there are other, less commonly known, orthoses that improve gait and gross motor skills in children with CP. To answer the overriding research question and to achieve these aims, one narrative systematic review and six studies were undertaken.

CP is described in depth throughout Chapter 1, further exploring the role of physiotherapy in paediatrics, specifically lower limb assessment and orthosis prescription. From assessment, physiotherapists are able to use their clinical reasoning and evidence-based research to prescribe therapy or aids (such as lower limb orthoses) to support their therapy management and achieve patient goals. Studies 1 and 2 described, and further investigated, current lower limb alignment assessment techniques, with particular focus on

techniques to ascertain subtalar joint neutral and the use of this measure to prescribe orthoses to treat tibial torsion.

Study 1 investigated the reliability of a lower limb alignment assessment technique. Fifteen adults and six children (n=21) were assessed in resting and neutral calcaneal stance position (RCSP and NCSP) through the Anterior Line Method by six allied health professionals. The allied health professionals were all familiar with the Anterior Line Method, with varying levels of experience from novice to expert. The results demonstrated that the RCSP appeared to be a more reliable measure than the NCSP across the ages. The findings of this study highlight that, the level of experience and familiarity of using the Anterior Line Method may influence reliability, as will measuring adult subjects as compared to children.

Using the records of the experienced assessor, the effect of orthotics with gait plates on tibial torsion in children was investigated. Through Study 2 (located in Appendix C: Tibial Torsion in Children: A Retrospective Study), chart data were assessed to determine if changes in tibial torsion occurred due to time or effect of orthotics. There was a final yield of 33 files for review. Within the reviewed charts, there were 23 male participants and 10 female participants (initial age = 2.5 -14 years). The mean period of time between initial assessment of RCSP and malleolar position (MP) and final assessment was 42.97 (± 41.41; range 3 – 147) months. Significant changes in RCSP and MP were found. As time was not a significant factor in the changes of left or right RCSP or MP, changes were likely due to the intervention or other unknown factors. However, a significant determinant of the amount of change was the initial torsion score. The results demonstrated there are physical improvements in tibial torsion which may be of benefit to the typical child through a simple, non-invasive method of intervention.

Due to the first two studies informing lower limb alignment assessment and orthoses in typical children only, these measures were therefore not considered appropriate for use in children with CP. The second study was relocated to Appendix C: Tibial Torsion in Children: A Retrospective Study, to maintain the total thesis journey, but so as to not interrupt the flow of the thesis. As such, further investigation into appropriate assessments and lower limb orthoses for children with CP was required to inform future studies.

One primary focus of this thesis was lower limb orthoses, such AFOs and SMotOs, and their effect on functional movement in this specialised population. To inform this focus, a narrative systematic review (Chapter 3: Study 3) was conducted into lower limb orthoses and their effect on gait and gross motor skills (GMS) in children with CP. Seven studies, graded as of good to high quality (Kennelly's grading system: 2011) informed the review.

The volume of evidence presented in the literature supported the use of AFOs as a device to improve gait and GMS in children with CP. However, there were inconsistencies in the agreement of design and in their use. A potential benefit of a clearer defining of AFO prescription within research studies was noted. In addition, given its lack, further research on the effect of SMotOs on gait and GMS in children with CP was identified as potentially being of value with regard to future orthoses prescription. Following the narrative systematic review, Chapters 4 – 7 presented further studies that explored AFOs and SMotOs and their impact on gait, GMS and quality of life in children with CP.

From the literature, as reported in Chapter 3 and through further reading, there appeared to be several clinically viable outcome measures to inform on changes in gait, GMS and quality of life in children with CP. These outcome measures were the Gross Motor Function Measure (GMFM-88), Berg Balance Scale (BBS), Timed Up-and-Go (TUG), Edinburgh Visual Gait Score (EVGS) and Cerebral Palsy Quality of Life (CPQoL) and it was these outcome measures that were selected as outcome measures to inform Study 4, a pilot feasibility study (located in Chapter 4).

The aim of the feasibility study was to determine the most effective and timely outcome measures of gait, GMS and quality of life in children with CP. Five outcome measures (GMFM-88, BBS, TUG, EVGS, CPQoL) were applied to six children with CP. A pre-determined minimum participation rate of 50% was set as the requirement for the outcome measure to be considered for future studies. Three outcome measures (GMFM-88, BBS and EVGS) achieved 50% or more participation rate.

Using the three predetermined outcome measures identified in Study 4 (Chapter 4), Studies 5 and 6 (Chapters 5 and 6) further investigated the effect of AFOs and SMotOs on GMS and gait in children with CP. Study 5 used the GMFM-88 and the BBS to determine the effect of SMotOs and AFOs on GMS in children with CP. Nine children (n=9: mean age= 5.4 ± 3.2 years: range 3–13 years), who wore both orthoses, were recruited via convenience sampling. Participants wore two different types of AFOs, being hinged-AFOs (n=4) and solid-AFOs (n=5). The GMFM-88 demonstrated medium positive change in three participants between orthoses. Sections D and E of the GMFM-88 demonstrated a 6% increase in score (SMotOs over AFOs). On average, participants scored significantly (p=0.002) higher on the GMFM-88 when wearing SMotOs compared to AFOs. There was no significant difference between BBS total scores when wearing SMotOs and AFOs (p=0.928). Using the GMFM-88, an effective and significant benefit on GMS was seen when wearing SMotOs compared to wearing AFOs.

To inform on changes in gait, and as identified in Study 4 (Chapter 4), the EVGS was deemed to be a clinically relevant, timely and effective outcome measure of gait changes in children with CP. Therefore, the EVGS was used to inform on the effect of SMotO and AFO on gait in Study 6. Eleven participants were videoed walking 5m (any order) barefoot, in SMotOs, and in AFOs. Of the participants (mean age = 5.5 ± 2.9 years: range 3 – 13 years) two (n=2) were female and six (n=6) used assistive devices. Seven (n=7) could walk barefoot. Participants presented with spastic diplegia (n=4), spastic quadriplegia (n=6), and spastic dystonic quadriplegia (n=1). Gross Motor Functional Classification System levels ranged I - IV. Total EVGS for SMotOs (7.62) and AFOs (14.18) demonstrated improved gait when wearing SMotOs with no significant differences between barefoot and AFOs. The results suggested SMotOs may be a viable option to improve gait in this population.

Along with the effect on gait and gross motor skills, a further interest was taken in how the orthoses affected the quality of life in children with CP. As the CPQoL did not meet the minimum participation rate (as per Study 4: Chapter 4), a specific quality of life questionnaire was created with the aim to identify key themes in relation to AFO and SMotO. A self-administered 24-item questionnaire was designed and consisted of two sections: section one related to AFOs and section two related to SMotOs. Study 7 (Chapter 7) employed a qualitative phenomenological approach to determine the effect of SMotO and AFO on quality of life. Participants were recruited via convenience sampling. Sixteen (69.57%) local and interstate families consented and returned the questionnaire. Of these, 81.3% were prescribed solid-AFOs, 18.8% were prescribed hinged-AFOs and one child was prescribed a dynamic-AFO (supramalleolar). There were four key themes identified: 1) time, 2) reason, 3) function, and 4) comfort and dislike. Half of the participants (50%) reported wearing both orthoses, 43.8% did not wear AFOs anymore and one wore AFOs only. The most common reason for AFO prescription was ‘alignment’ (43.8%). Six families (37.5%) were told the reason for AFOs being prescribed to their child was ‘because the child has CP.’ There were numerous functional improvements seen in both AFOs and SMotOs, such as walking and balance. When asked if their child was comfortable wearing their AFOs, 43.8% reported a mix of comfortable and uncomfortable, and fifteen families (93.8%) disliked the restriction in movement from the AFOs. Ten families (62.5%) disliked the way the AFOs fit into shoes, and nine families (56.3%) disliked the bulkiness of the AFOs. With regard to SMotO, five families (31.3%) disliked the way they fit into shoes and seven families (43.8%) reported their child experienced pressure areas. Overall, the results trended towards a preference of wearing SMotOs for comfort, function, and in general, as preference to AFOs.

Having investigated both quantitative and qualitative data from the participants, to further create depth to the body of research presented, a case series was conducted with the specific focus to provide snapshots of the individual children with CP. Chapter 8 presented a case study series aimed to enrich the volume of work presented in this thesis, and to provide a clinically relevant picture to physiotherapists working with children with CP.

Overall, this program of research demonstrated the Anterior Line Method and gait plate orthoses may not be viable for children with CP and established that there is a significant positive effect on gait, gross motor skills, and quality of life in children with CP when they wear SMotOs when compared to AFOs. Clinically, SMotOs provide an alternate option for AFOs in children with CP and as such, with further research, could become a wider used orthoses in this population. Future research should include investigations into creating clinically applicable lower limb alignment assessment techniques, modifications to current AFOs and further expand evidence supporting the use of SMotO to improve gait, GMS and quality of life in children with CP.

Key Words

paediatrics; ankle-foot orthoses; sensomotoric orthoses; cerebral palsy; gross motor skills; GMFM-88; Berg Balance Scale; gait; Edinburgh Visual Gait Score; quality of life; tibial torsion; anterior line method; lower limb alignment

Declaration by Author

I certify that this thesis document submitted to Bond University is in fulfilment of the requirements for a Doctor of Philosophy (PhD). This thesis represents my own original work towards this research degree and contains no material which has been previously submitted for a degree at this University or any other institution, except where due acknowledgement is made.

Clare MacFarlane

20th April 2020

Declaration of Author Contributions

All author contributions to manuscripts under review have been listed below in Table 1. These authors have provided approval for publication within this thesis.

Table 1: Manuscript author contributions

Manuscript	Contribution
MacFarlane C, Simas V, Hing W, Orr R. Using the Edinburgh Visual Gait Score to Compare Ankle-Foot Orthoses, Sensorimotor Orthoses and Barefoot Gait Pattern in Children with Cerebral Palsy. (Published, *Children (2020)7(6):54*).	CM 70%, VS 10%, RO 10%, WH 10%
MacFarlane C, Simas V, Hing W, Orr R. Orthoses, Gait, Gross Motor Skills and Children with Cerebral Palsy: A Systematic Review. (Submitted for publication).	CM 70%, VS 10%, RO 10%, WH 10%
MacFarlane C, Simas V, Hing W, Orr R. Effect of Sensomotoric Orthoses and Ankle-Foot Orthoses on Gross Motor Skills in Children with Cerebral Palsy. (Submitted for publication).	CM 70%, VS 10%, RO 10%, WH 10%
MacFarlane C, Orr R, Hing W. Sensomotoric Orthoses, Ankle-Foot Orthoses and Children with Cerebral Palsy Case Series: The Bigger Picture. (Published, *Children (2020)7(8):82*).	CM 80%, RO 15%, WH 5%
MacFarlane C, Hing W. Effect of Sensomotoric Orthoses and Ankle-Foot Orthoses on Quality of Life in Children with Cerebral Palsy. (Submitted for publication).	CM 85%, WH 15%

Research Outputs

Peer reviewed publications

- **MacFarlane, C.**, Simas, V., Hing, W., Orr, R. (2020). Using the Edinburgh Visual Gait Score to Compare Ankle-Foot Orthoses, Sensomotoric Orthoses and Barefoot Gait Pattern in Children with Cerebral Palsy. *Children*, 7(6): 54.
- **MacFarlane, C.**, Orr, R., Hing, W. (2020). Sensomotoric Orthoses, Ankle-Foot Orthoses and Children with Cerebral Palsy Case Series: The Bigger Picture. *Children*, 7(8): 82.

Manuscripts Submitted for Publication

- **MacFarlane, C.**, Simas, V., Hing, W., Orr, R. Orthoses, Gait, Gross Motor Skills and Children with Cerebral Palsy: A Systematic Review.
- **MacFarlane, C.**, Simas, V., Hing, W., Orr, R. Effect of Sensomotoric Orthoses and Ankle-Foot Orthoses on Gross Motor Skills in Children with Cerebral Palsy.
- **MacFarlane, C.**, Hing, W. The Effect of Sensomotoric Orthoses and Ankle-Foot Orthoses on Quality of Life in Children with Cerebral Palsy.

Ethics Declaration

In order to ensure that the proposed studies were conducted in accordance with relevant ethical standards and guidelines and to ensure protection of the welfare and rights of participants in this research, all proposed studies were submitted to the Bond University Human Research Ethics Committee (BUHREC) The research associated with this thesis received ethics approval from BUHREC with relevant approvals listed in Table 2 below.

Table 2: Ethics application status

Application	Ethics Committee	Status
EC-00357	BUHREC	Approved
RO-1539	BUHREC	Approved
RO-1835	BUHREC	Approved

Copyright Declaration

This thesis makes careful note of all sections which have been previously published, along with relevant copyright material.

There was no copyright permission required as the two manuscripts accepted for publication were by the Open Access Journal *Children*.

1. MacFarlane, C., Simas, V., Hing, W., Orr, R. Using the Edinburgh Visual Gait Score to Compare Ankle-Foot Orthoses, Sensomotoric Orthoses and Barefoot Gait Pattern in Children with Cerebral Palsy. *Children* 7(6), DOI: 10.3390/children7060054. Under Creative Commons licence CC BY 4.0.

2. MacFarlane, C., Orr, R., Hing, W. Sensomotoric Orthoses, Ankle-Foot Orthoses and Children with Cerebral Palsy Case Series: The Bigger Picture. *Children*, 7(8), DOI: 10.3390/children7080082. Under Creative Commons licence CC BY 4.0.

Inspiration

This thesis body of research has been fuelled by my experience in various paediatric private practice settings. Since completing my Doctor of Physiotherapy in 2011, I have continued to develop my skills in paediatric physiotherapy and, over the years, I have learned the importance and value of working alongside other allied health professionals. This has included podiatrists, occupational therapists, speech therapists and osteopaths. I believe taking a multi-disciplinary approach to therapy for children is the most effective process for working holistically with the body and its needs. Throughout my experience in various clinical settings, I became fascinated at the role orthotics played in alignment and how they could assist the children I treated.

Through working alongside podiatrists, I learned different assessment techniques and prescription options for orthotics in both adults and children. This then led me to investigate the reliability of previously unresearched assessment techniques, the effectiveness of orthotics in changing tibial torsion and the impact of specialised orthotics on gait and gross motor skills in children with cerebral palsy (CP).

Throughout my role as a Paediatric Physiotherapist, Director, and the Director of Rehabilitation at Neurological and Physical Abilitation (NAPA) Centre in Sydney, I have provided many aspects of therapy that demonstrates progression toward better client and community health care outcomes. This is a specialised private practice clinic of paediatric physiotherapists, occupational therapists and speech pathologists, working collaboratively within the team and alongside podiatrists to provide intensive rehabilitation therapy programs for children with significant disabilities, such as (but not limited to) CP, global developmental delays, autism, down syndrome, acquired brain injuries, genetic and metabolic disorders, and other neurological conditions.

Completing this thesis has been a humbling experience. Working closely with families and hearing their stories from a different perspective has created a new appreciation of the difficulties that these complex special needs families experience. A driving force in my doctoral studies has been my hope that the findings of my work can be used to assist families and therapists to determine the best course of action and therapy approach for their child or client through uncovering and exploring therapeutic options for children with CP. Now that I have completed my research, I hope to fulfil this goal.

As clinical physiotherapists, we need to objectively assess and look at the use and provision of orthoses in this population. The following chapters serve to provide theoretical and practical options, through research, to enhance paediatric physiotherapy evidence-based practice in children with CP.

- Clare MacFarlane

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It is with heartfelt gratitude and honour that I recognise the incredible guidance and support provided by my supervisors throughout the journey, and subsequent development, of this thesis manuscript.

To Professor Wayne Hing, my primary supervisor, your encouragement from the start, never allowing me to lose faith in myself or the process has allowed the creation of these words. Your constant guidance, motivation and problem-solving skills has uplifted me and created resilience I didn't know existed, all whilst being in absolute awe of your knowledge! Thank you.

To Dr. Rob Orr, my secondary supervisor, your incredible editing and going above and beyond to provide feedback over weekends (and your time off) has had me humbled by both your work ethic and unbelievable knowledge. Your commitment to this thesis and its fine tuning has been remarkable, an overwhelming thankyou is merited.

To Dr. Vini Simas, my co-author, editor and supervisor, your knowledge and assistance has been incredible and invaluable. Your patience, support and clarity in explanation made this whole process a lot more manageable and calmer. I appreciate your guidance, thank you!

A huge thank you to the three clinics (ICB Gait & Posture, Therapies for Kids and NAPA Centre) and participating clinicians for allowing the data collection and research to be undertaken, your generosity and support has been key. All of which would not have been possible without all my amazing participants, thank you for choosing to be a part of this research!

To my wonderful husband, Brenton, whose gentle words and care has lifted me through the toughest times, thank you for believing in me, and supporting me through these years! I promise our future years will be void of thesis work and full of adventures.

Last, but far from least, to my parents, whose support and belief in me has boosted me through every little and big challenge this thesis (and life) has thrown at me – thank you from the bottom of my heart.

Thank you, thank you, and thank you again!

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List of Abbreviations

Ankle-Foot Orthoses	AFO
Anterior Line Method	ALM
Analysis of Variance	ANOVA
Australian Physiotherapy Association	APA
Active Range of Motion	AROM
Berg Balance Scale	BBS
Bruininks-Osteresky Test of Motor Proficiency	BOTMP
Bond University Human Research Ethics Committee	BUHREC
Critical Appraisal Score	CAS
Cerebral Palsy	CP
Cerebral Palsy Quality of Life Questionnaire	CPQoL
Computed Tomography	CT
Dynamic Ankle Foot Orthoses	DAFO
Developmental Dysplasia of the Hip	DDH
Evidence-Based Medicine Levels of Evidence	EBMLE
Ethyl Vinyl Acetate	EVA
Edinburgh Visual Gait Score	EVGS
Foot Posture Index	FPI
Gross Motor Ability Estimator	GMAE
Gross Motor Function Classification System	GMFCS
Gross Motor Function Measure (-88 or -66)	GMFM
Gross Motor Performance Measure	GMPM
Gross Motor Skills	GMS
Gait plate	GP
Hinged Ankle-Foot Orthoses	HAFO
Confidence Interval	IC
Intraclass Coefficient	ICC
Hinged Ankle Foot Orthosis	HAFO
Left	(L)
Lower Limb Orthoses	LLO
Modified Ashworth Scale	MAS
Minimal Clinically Important Difference	MCID

Malleolar Position	MP
Neutral Calcaneal Stance Position	NCSP
Outcome Measure	OM
Postural Control Insoles	PCI
Pediatric Evaluation of Disability Inventory	PEDI
Posterior Leaf Spring Orthoses	PLS
Pediatric Outcomes Data Collection Instrument	PODCI
Preferred Reporting Items for Systematic Reviews	PRISMA
Passive Range of Motion	PROM
Prospectively Registered Systematic Reviews in Health and Social Care	PROSPERO
Questionnaire	Q'AIRE
Quality of Life	QoL
Resting Calcaneal Stance Position	RCSP
Randomised Controlled Trial	RCT
Range of Motion	ROM
Right	(R)
Solid Ankle-Foot Orthosis	SAFO
Standard Deviation	SD
Sensomotoric Orthoses	SMotO
Statistical Package for the Social Sciences	SPSS
Subtalar Joint Neutral	STJN
Tibial counter rotator	TCR
Tibial Torsion	TT
Timed Up-and-Go	TUG
Initial assessment	T1
Final assessment	T2

(Abbreviations will be in full at the start of each chapter)

Thesis Structure

The overarching aim of this program of research is to explore the assessment and prescription of lower limb orthoses in the paediatric cerebral palsy (CP) population and how these orthoses affect gait, gross motor skills and quality of life. This was addressed through six objectives over nine chapters.

Chapter 1 provides background context to CP and the role of paediatric physiotherapy. This chapter describes the general outline of what CP is and implications for the lifestyle of a child. Chapter 1 includes a contextual description of how the project ideas evolved as well as an introduction to physiotherapy and the general areas a physiotherapist can specialise in, with particular focus on the role of a paediatric physiotherapist.

Chapter 2 reviews the methods of lower limb assessment through a proof-of-concept experimental design study, focussing on the reliability of the Anterior Line Method. The Anterior Line Method has been used to assess subtalar joint neutral and prescribe orthotics in both the adult and paediatric population. The aim of the proof-of-concept design was to assess the reliability and applicability to a paediatric population. Due to lack of reliability, the Anterior Line Method was not implemented in further studies as originally planned. This study is followed by a review of lower limb alignment, specifically the concept of tibial torsion, assessment, measurements and treatment options, thus leading into a retrospective chart review (Appendix C) that investigates the effect of orthoses on tibial torsion in children over time.

To further investigate lower limb orthoses, **Chapter 3** reviews lower limb orthoses in general, with specific details surrounding ankle-foot orthoses (AFOs) and sensomotoric orthoses (SMotOs) in children with CP. This leads to a narrative systematic review of the effect of lower limb orthoses on gait and gross motor skills in children with CP.

When assessing children with CP, there are several outcome measures that determine gross motor skills and gait. **Chapter 4** presents a feasibility pilot study to evaluate the most timely and effective outcome measures to assess children with CP in two different types of orthoses (AFO and SMotO) in preparation for further in-depth investigation presented in the following two sibling chapters, Chapter 5 and Chapter 6.

Chapter 5 presents a study investigating the effects of SMotOs and AFOs on gross motor skills in children with CP, through implementation of the selected timely and effective outcome measures as per Chapter 4.

To continue the theme of gross motor skills, **Chapter 6** further refines gross motor skills to investigating the effects of SMotOs and AFOs on gait in children with CP. The effects on gait are found through the Edinburgh Visual Gait Score, as per Chapter 4's outcome measure feasibility study.

Further to support the in-depth examination of the effect of lower limb orthoses in children with CP, **Chapter 7** presents a qualitative analysis investigating the effect of AFOs and SMotOs on quality of life for children with CP through a parent centred questionnaire. To describe the 'bigger picture,' **Chapter 8** continues this theme by a case series presenting the effect of AFOs and SMotOs on quality of life, gait and gross motor skills in children with CP.

Chapter 9 closes the studies and case series, summarising all study results, acknowledging the limitations of the thesis, and outlining the clinical relevance of findings. It concludes with recommendations for future research. Please see Figure 1 for overview of the body of research involved in this thesis.

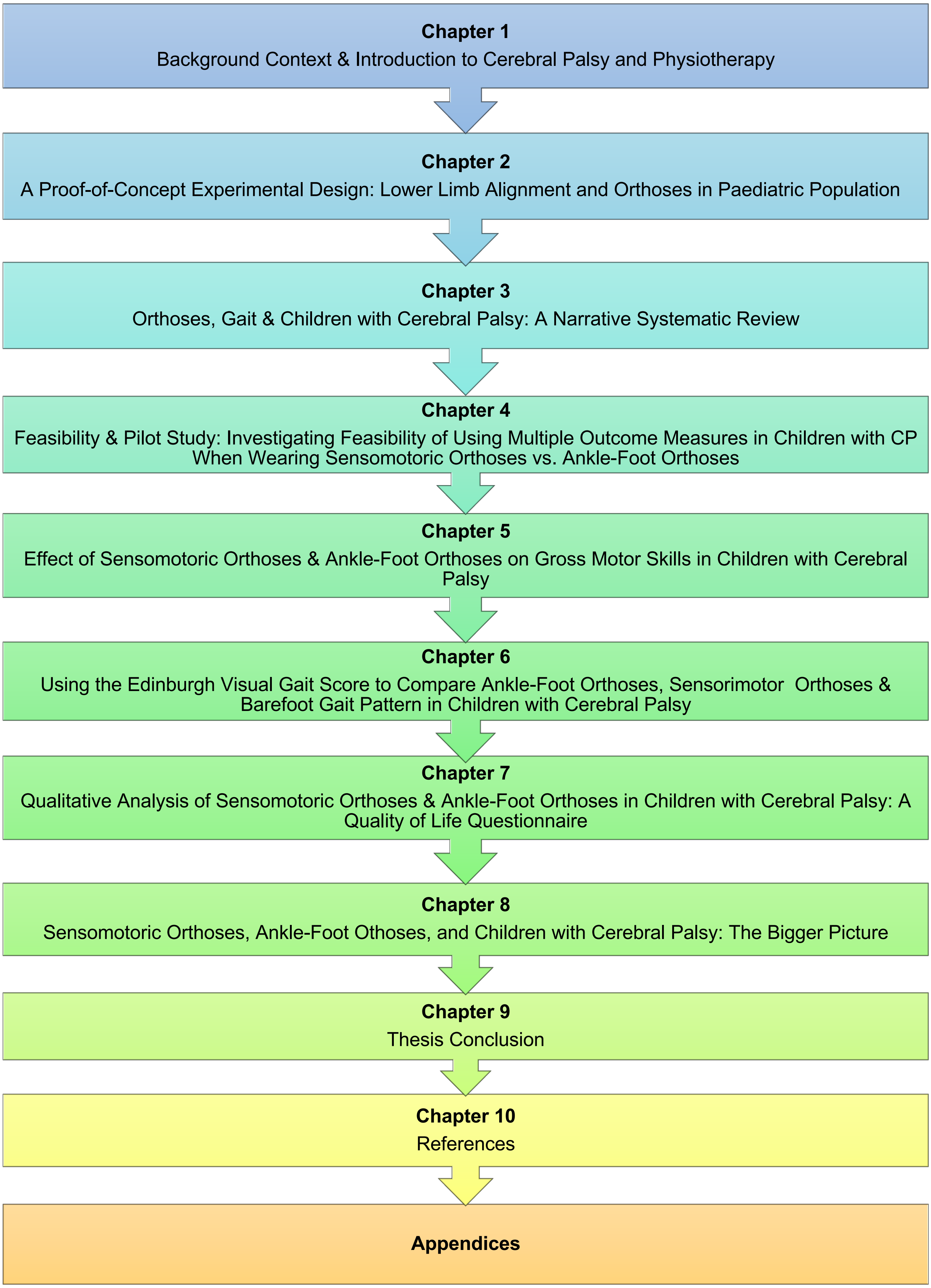


Figure 1: Overview of the program of research presented in this thesis

Research Aims & Hypotheses

Of the current literature in the field of paediatric physiotherapy, no studies have investigated the effects of sensomotoric orthoses on gait, gross motor skills, and quality of life in children with CP. This thesis examined, in detail, clinical assessment techniques, usual outcome measures for gait and gross motor skills, and outlined commonly prescribed orthoses in children with CP.

The overarching aim of this program of research was to answer the research question 'Which lower limb orthoses are optimal for treating children with CP?' through exploring the assessment and prescription of orthoses in a paediatric population and how these affected gait, gross motor skills and quality of life in children with CP. Specific aims and hypotheses included:

1) *To describe and outline the background of physiotherapy and cerebral palsy, providing context for paediatric physiotherapy and how it supports those children with cerebral palsy.*
2) *To investigate through proof-of-concept experimental design, focussing on the reliability of a lower limb assessment technique, the Anterior Line Method (ALM), specific to the paediatric population. It was hypothesised that the ALM would be more reliable when implemented by more experienced assessors. Through further exploration and evaluation, the implementation of orthoses in a paediatric setting to address tibial torsion will be investigated.*
3) *Systematically review the literature with the focus on children with cerebral palsy and orthoses, and what effect lower limb orthoses have on improving gait and gross motor skills. It was hypothesised that lower limb orthoses would improve alignment, cadence, and balance.*
4) *Explore the feasibility of several outcome measures to be used in a timely and effective manner with the intent of determining the effect of two types of orthoses (sensomotoric and ankle-foot) on gait and gross motor skills involving children with cerebral palsy, using gross motor measures and the Edinburgh Visual Gait Score. It was hypothesised that three outcome measures would be feasible to implement in a*

timely manner and that gait and GMS would be improved when wearing SMotOs more than AFOs.

5) *To investigate the effect of lower limb orthoses (AFOs and SMotOs) on gait and gross motor skills in children with CP. It was hypothesised that the SMotO would demonstrate better results in both gross motor skills and gait, with AFOs demonstrating better results in static balance.*
6) *Display and compare the qualitative analyses surrounding the benefit of two types of orthoses on the quality of life through parental feedback and a case series of children with cerebral palsy, especially relating to gait and gross motor skills. It was hypothesised that families would prefer the SMotOs over the AFOs for over 50% of the feedback.*

1.0. Background Context and Introduction to Cerebral Palsy and Physiotherapy

Prelude

The following chapter introduces the condition of Cerebral Palsy (CP) as a physical disability and goes on to summarise and describe the motor types and the classification from a gross motor aspect. The chapter then describes physiotherapy with involvement relating to the role of the paediatric physiotherapist in the treatment and intervention of the paediatric CP population. This chapter also explores how therapists support the use of orthoses for mobility and the subsequent effect on quality of life, with a specific focus on how the concept for this program of research originated.

1.1. Cerebral Palsy

Cerebral Palsy (CP) is a neurodevelopmental condition well recognised to begin at birth or early childhood and persisting through the lifespan and is the leading cause of physical disability in children (Paulson & Vargus-Adams, 2017; Rosenbaum, Paneth, Leviton, Goldstein, Bax, 2007). CP describes a group of permanent, non-progressive disorders affecting the development of movement and postures and causing activity limitation (Christovao et al., 2015; Ridgewell, Dobson, Bach, & Baker, 2010; Rosenbaum et al., 2007). The primary cause of CP is damage to the developing brain, which can cause secondary alterations in the locomotor apparatus (Romkes & Brunner, 2002) such as muscle spasticity.

CP is the most common permanent physical disability in childhood (Paulson & Vargus-Adams, 2017) with an incidence of 2.0 to 2.5 per 1000 live births. It is characterised by abnormal motor patterns and postures, with a variety of presentations (Graham & Selber, 2003), such as spasticity, dystonia, muscle contractures, weakness and difficulty in co-ordination, ultimately affecting control of movements (Paulson & Vargus-Adams, 2017).

CP is often accompanied by disturbances of sensation, perception, cognition, communication, as well as eating and drinking, and behaviour, by epilepsy and by secondary musculoskeletal problems (Paulson & Vargus-Adams, 2017; Rosenbaum et al., 2007).

1.2. Topographical Distribution (Types)

According to the Cerebral Palsy Alliance, NSW (2020), there are three types of CP:

- *Quadriplegia*: where all four limbs are affected, as well as muscles in the trunk, face and mouth often affected.
- *Diplegia*: where both legs are affected.
- *Hemiplegia*: where one arm and leg on the same side are affected.

Quadriplegic CP can also be described as bilateral CP and hemiplegic CP can be described as unilateral CP (Cans et al., 2007). The description of the topographical CP description can be further added to by including motor type and severity.

1.3. Motor Types of CP

These types are further classified into presentation (Cans et al., 2007) and can be further defined through following the classification tree (Figure 2):

- *Spastic*: damage in the motor cortex affecting 70-80% of individuals (most common form), increased tone and spasticity with pathological reflexes.
- *Dyskinetic*: damage in the basal ganglia affecting 6% of individuals, demonstrated by uncontrolled, involuntary, recurring movements with primitive reflex patters and varying muscle tone.
- *Ataxic:* damage in the cerebellum affecting 6% of individuals and is characterised by shaky movements due to the loss of orderly muscle co-ordination, compromising balance and proprioception with a predominance of low tone.
- *Mixed*: combination of the damages described above. Child is classified further according to the dominant clinical feature.

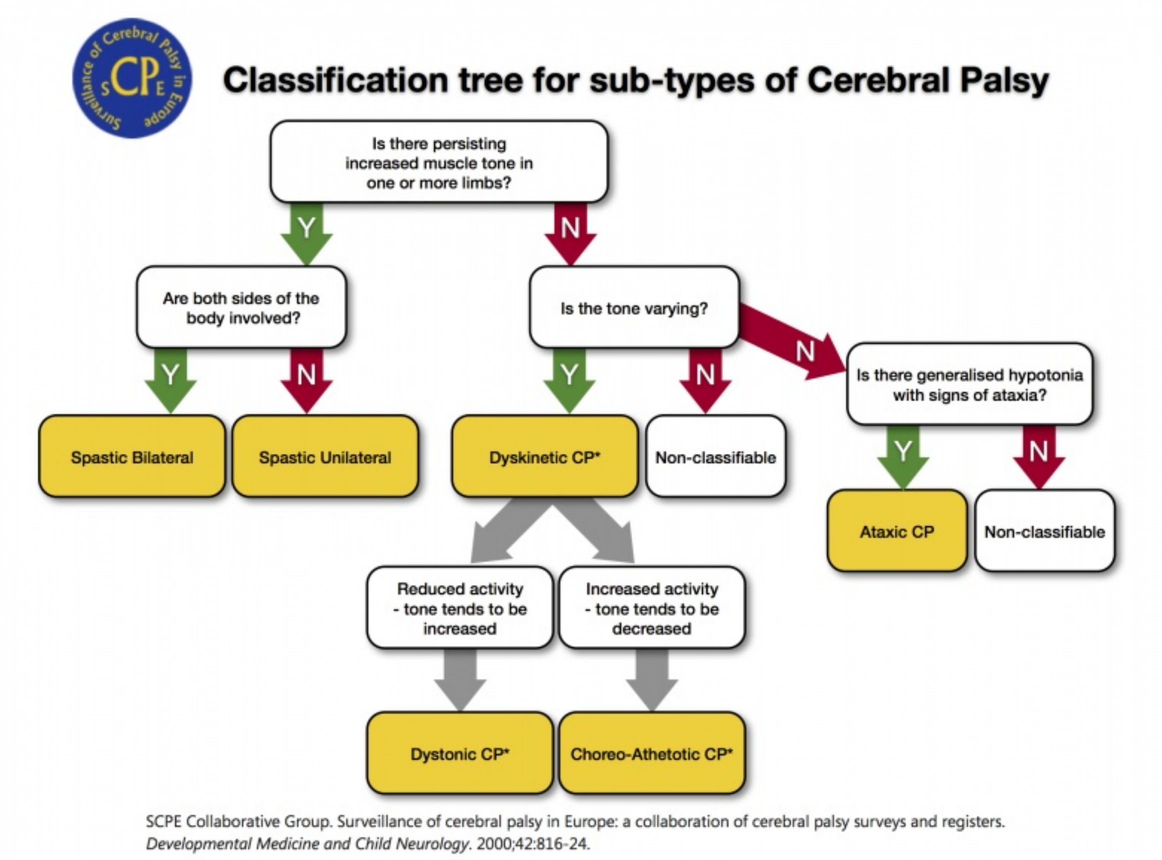


Figure 2: Classification of CP sub-types (Cans et al., 2007)

1.4. Severity and Classification

Generally, severity can be classified into mild, moderate and severe. To further identify the areas affected by severity, the effect of the child's CP can be classified by four different systems (Paulson & Vargus-Adams, 2017).

- *Gross Motor Function Classification System (GMFCS):* classifies the child's movements such as sitting, walking, and use of mobility devices. Most established and recognised of the functional classification systems (Paulson & Vargus-Adams, 2017).
- *Communication Function Classification System:* classifies the child's communication according to effectiveness, into one category of five (Paulson & Vargus-Adams, 2017).
- *Manual Ability Classification System:* classifies the child into one of five categories for fine motor and handling skills (Paulson & Vargus-Adams, 2017).
- *Eating and Drinking Ability Classification System:* valid measure to assess eating and drinking ability for children with CP (3-years and older) (Paulson & Vargus-Adams, 2017).

This thesis will only refer to and use GMFCS levels as it is the most relevant to the content of the studies undertaken. All other classifications are with regard to communication, fine motor skills and feeding ability and therefore not relevant to orthoses and lower limb in children with CP.

1.5. GMFCS Classification

The GMFCS describes the function of a child through self-initiated movement and the use of assistive mobility devices. It has a simple five-level grading system that is used to describe the function of the child with CP, starting at GMFCS-I being able to walk without limitations to GMFCS-V requiring full assistance in a wheelchair.

The GMFCS is the most established and recognised of the functional classification measures in CP (Paulson & Vargus-Adams, 2017). It was developed in 1997 and found to be reliable (interrater reliability k=0.55 for below 2 years old and k=0.75 for those aged 2-12 years) amongst 48 experts (Palisano et al., 1997). Following research to stratify typical motor function for children with CP, the authors concluded that a five-level classification system worked well to discriminate clinically meaningful distinctions in motor function. This classification system was originally designed for children with CP from 2–12 years of age (Palisano et al., 1997). The GMFCS was later expanded and revised in 2007 to include ages 12–18, as well as to increase descriptors and differentiations for the levels based on the child's age, while taking into account developmental milestones (Paulson & Vargus-Adams, 2017).

GMFCS Levels based on the revised and expanded version (Paulson & Vargus-Adams, 2017).

- I - walk without limitations:
    - >2 years old: crawl on hands and knees, pull to stand, cruise along furniture and independent walking (between 18-24months)
    - 2-4 years: independent sitting and independent transitions to standing
    - 4-6 years: walk independently indoors and outdoors, climb stairs, begin to run and jump
    - 6-18 years: walking up and down curbs, ambulating in the community, climb stairs without railings and run and jump
- II - walk with limitations:
    - >2 years old: sit with upper extremity support, crawl on tummy, may be able to pull to stand with support
    - 2-4 years: transitions into and out of sitting, sit without support (may need to use upper extremities), crawl on hands and knees, cruise with support and mobilise with device
    - 4-6 years: transition in and out of standing, walk short flat distances, climb stairs with railing, unable to run or jump
    - 6-18 years: walk in most terrains but has limitations distance or uneven surfaces, may use wheeled or handheld devices for long distances, climb stairs with raining, minimal or no running and jumping
- III – walk with handheld device indoors, but wheeled device outdoors or longer distances:
    - >2 years old: roll and occasionally crawl on stomach, sit with some low back support
    - 2-4 years: 'W' sit on floor with some assistance to transition, crawl on stomach or crawl on hands and knees, may pull to stand and walk short distances with walker or gait trainer and some assistance for manoeuvring
    - 4-6 years: sit in a standard chair but may require extra support for upper limb use, walk with handheld device, climb stairs with assistance, wheeled mobility for longer distance

    - 6-18 years: mobilise with handheld device indoors, wheeled mobility for distance, assistance to transition into sitting and standing, negotiates stairs with assistance
- IV – limited self-mobility, usually transported in a wheelchair:
    - >2 years old: head control and can roll, require truncal support to sit
    - 2-4 years: sit with upper extremity support, assistance to transition into sitting, may require adaptive equipment for sitting or standing, some self-mobility possible through rolling or stomach crawling short distances without reciprocal leg movement
    - 4-6 years: require adaptive equipment to control trunk to allow sitting, assistance to transition between positions, may walk short distance with device and assistance, wheeled mobility for distances
    - 6-18 years: require adapted seating, assistance with transfers, utilise wheelchair with assistance in most settings, can have independent floor mobility with crawling or rolling, or may walk short distances with assistance
- V – severe limitations
    - >2 years old: no independent head or trunk control, assistance to roll
    - 2-4 years: no independent movement, requires assistance for transport using manual mobility devices, require adapted equipment for sitting and standing but function is still limited, may be possible to become independent using power mobility
    - 4 onwards: abilities are stable with the need for complete assistance with transfers emerging after age 6

1.6. Physiotherapy and Role of Paediatric Physiotherapy

The Australian Physiotherapy Association (APA), the governing body for physiotherapists, describes physiotherapists as *'highly qualified health professionals who, through evidence-based practice, can assess, diagnose and treat many conditions relating to health and physical disorders, thus leading to an increase in mobility and improve quality of life.'*

There are many specialist areas of physiotherapy that a potential patient can access through both the public and private health care system. Alongside musculoskeletal or sports physiotherapy, other physiotherapy specialities, as described on the APA website (APA 2017), include:

- Cardiorespiratory: prevents disease and rehabilitates those with diseases and injuries affecting the heart and lungs
- Cancer, palliative care and lymphoedema: supports patients through managing or preventing fatigue, pain, muscle and joint stiffness, and deconditioning
- Continence and women's health: manage and prevents incontinence and pelvic floor dysfunction in men, women and children
- Geriatric: promoting healthy and active ageing among older Australians
- Neurology: promotes movement and quality of life in patients who have had severe brain or spinal cord damage from trauma, stroke, or neurological conditions
- Orthopaedic: prevent or manage acute or chronic orthopaedic conditions

One key area of physiotherapy, and the one that is relevant to this research, is paediatric physiotherapy, which can be accessed in the public, private or non-profit government sectors of health care. The APA (2013) describes paediatric physiotherapists as having '*an in-depth knowledge of all aspects of development, and have the clinical assessment, reasoning and intervention skills for best practice management of acute, non-acute, lifelong and life limiting conditions*.'

Paediatric physiotherapists provide population-specific therapy or treatment for those aged between 0-18 years, by reducing or preventing the onset of secondary conditions by optimising mobility and strength or support a child's development such as addressing milestone delays with sitting and walking or clumsiness (APA 2008). The physiotherapist supports the child through therapeutic handling, specific positioning, educating parents on how to perform this therapy at home through fun games aimed at strengthening and skill acquisition. Some children may require equipment to support them in achieving goals of physical development, such as walking frames, standing frames or orthoses. Physiotherapists play an important role in prescribing aids and equipment (APA 2006) to aid with movement, to create improved body alignment and to assist children to integrate this equipment use (APA 2008).

Other physical measures available to physiotherapists include observational gait assessments, gross motor skills assessments, such as the Gross Motor Measure of Function (GMFM-88 or -66), Timed Up and Go (TUG), Berg Balance Scale (BBS) and other types of evaluation to fully understand the function of their client/patient. Evaluations can include lower limb alignment assessment, such as range of motion or position of lower limbs.

This is further outlined in Chapter 2, whereby it discusses accuracy and reliability as well as outlining gold standard assessments.

Current evidence-based practice demonstrates the effectiveness of a multi-disciplinary intervention. In a systematic review and pilot analysis, Craig (1999) notes that children with CP are now presenting with more severe and more complex problems, which necessitate multi-dimensional intervention. Such concept requires the ability to communicate and coordinate treatment with other allied health professionals. This ensures a holistic treatment and maximises the skillset of all health professionals involved in the treatment. Chan et al. (2010) highlights the benefits that such approach has for organisations, teams, patients and clinicians include reduction in hospitalisation, improvement in care coordination, better health outcomes and job satisfaction.

Furthermore, Craig (1999) concluded her paper by reporting that '*client centred practice, interdisciplinary teamwork and evidence-based practice have become familiar words in the vocabulary of the health profession. Therapy application is complex, as the review of literature within this study has reinforced. Physiotherapy has been indicated through this research as a core discipline in the management of children with a disability such as cerebral palsy, making the application of such theory to practice a necessity.*'

1.7. Intervention & Orthoses for CP

CP is the main neurological referral within paediatric physiotherapy (Craig, 1999). In an editorial by Perat (2012), it is noted that although there is not one best treatment for all problems associated with CP, there is no doubt that if treatment starts early on, then better results will be achieved. This is further explained to include early detection assists with adequate intervention. Physically, in children with CP, neuromuscular impairments such as spasticity, reduced motor control and proprioception, can affect the typical, co-ordinated movement required for gait (Danino et al., 2015). Gibson (2013) noted that the achievement of independent walking is a major focus of rehabilitation for children with CP, and that the perspectives and experiences of parents and children regarding walking and walking therapies remain largely absent from professional debates.

Motor impairment is the main manifestation in children with CP, with consequent alterations in the biomechanics of the body (Christovao et al., 2015). Ries (2017) describes spasticity as any motion of an agonist muscle being counteracted by the antagonist muscle as it is stretched. This elicits a spastic catch and is prohibitive to intentional movement.

Spasticity and muscle stiffness have an adverse effect on the normal formation of the skeletal system and its growth, resulting in bony or alignment deformities.

Along with physiotherapy intervention, lower extremity orthoses – such as ankle foot orthoses (AFOs) - are often used as a treatment to address the biomechanical limitations and joint alignment (Bjornson, Schmale, Adamczyk-Foster, & McLaughlin, 2006), as well as to promote functional activities (Figueiredo, Ferreira, Moreira, Kirkwood, & Fetters, 2008) for children with CP. AFOs support normal joint alignment and mechanics, provide variable range of motion (ROM) when appropriate, and facilitate function (Brodke et al., 1989; Knutson & Clark, 1991; White, Jenkins, Neace, Tylkowski, & Walker, 2002). AFOs are further explored and extensively described in Chapter 3.

AFOs, specifically, are employed within a paediatric physiotherapy approach for several treatment reasons, including stabilising the ankle / foot complex (Buckon, Jakobson-Huston, Moor, Sussman, & Aiona, 2004), enabling a continuous Achilles / gastrocnemius stretch (Boyd, Pliatsios, Starr, Wolfe, & Graham, 2000), to obtain heel strike during gait (Carmick, 2013) and prevent contracture of the Achilles / gastrocnemius developing (Hainsworth, Harrison, Sheldon, & Roussounis, 2007; Morris, 2002). Along with joint alignment, other improvements that may be seen with the use of AFOs are maintaining ankle ROM, stability of the ankle joint (Buckon et al., 2004; Smith et al., 2009), prevent equinus positioning, improve walking efficiency (Rethlefsen, Kay, Dennis, Forstein, & Tolo, 1999), balance (Carmick, 2013), and to improve gait function (Westberry et al., 2007). Although, in contrary, Carmick (2013) reported when foot orthoses block a critical movement (i.e., ‘foot rockers’), children compensated the lack of forefoot rocker (as in AFOs) by internally rotating their legs, walking in equinus, and falling. Usually made by an orthotist, AFOs are comprised of polypropylene that cover the entire posterior calf and the mediolateral borders and sole of the foot, with straps across the anterior upper tibia and front of the ankle (Radtka, Skinner, Dixon, & Johanson, 1997). Common styles, as reported by Wren et al. (2015) include solid-AFOs (SAFO) and hinged-AFOs (HAFO) with dorsiflexion or plantarflexion stops, as well as posterior leaf spring (PLS) orthoses (Figure 3) and dynamic-AFOs (DAFOs: Figure 4). SAFOs restrict dorsiflexion or plantarflexion and usually have arch support and mediolateral control built in. HAFOs allow for dorsiflexion but limit plantarflexion, whereas PLS are used for compensating weak ankle dorsiflexors and have no mediolateral control[1].

[1] For more information regarding AFOs please see Chapter 3: A Systematic Narrative Review.

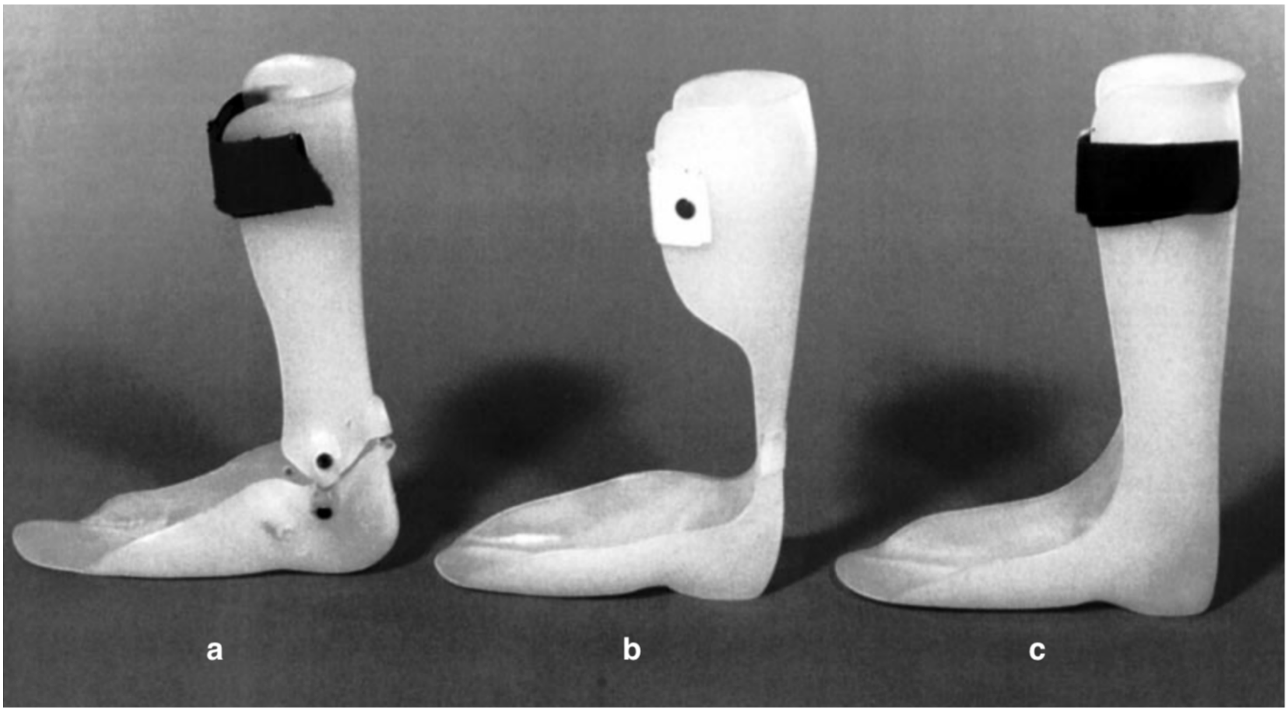


Figure 3: Ankle Foot Orthoses - Hinged (a), Posterior Leaf Spring (b) and Solid (c) (Buckon et al, 2001. Comparison of three ankle-foot orthosis configurations for children with spastic hemiplegia. Wiley Online Library. Copyright © 1999-2021 John Wiley & Sons, Inc. All rights reserved).

Figure 4: Dynamic AFO (cascadedafo.com/products/custom-dafo)

Unlike AFOs, sensomotoric orthoses (SMotOs)[2] provide a different approach to the management of gait and spasticity in children with CP. The SMotOs are created to activate and deactivate muscles by increasing or decreasing point specific pressure on musculotendinous structures in the foot. Jahrling (2001), the creator of the SMotOs, reported that the orthoses are fast-acting as they activate, deactivate and stabilise the foot and

[2] Figures of SMotO are provided later in this thesis. See Figures 11 & 12 in Chapter 6.

perform a ‘push and pull’ effect in the foot. The Golgi bodies (in the musculotendinous junctions of the tendons of the foot) are activated to relax or contract the targeted muscles by the pressure from the ‘bumps’ coinciding with the muscles (Jahrling, 2001). The theory lies in information being sent by afferent feedback and are thought to reduce the activity over over-active muscles through inhibition, and therefore facilitate an increase in the activity of weaker muscles (Ludwig, Quadflieg, & Koch, 2013). Ludwig, Kelm and Fröhlich (2016) investigated whether the activity of the peroneus longus muscle could be increased by the targeted use of a specially formed lateral pressure element in a sensomotoric insole. They found, in a laboratory-based randomised crossover design study using surface electromyography in 34 healthy participants, that in 27/34 participants an additional activity peak of the peroneus longus muscle was observed during the loading response phase and mid-stance phase. They concluded that the pressure point changes afferent information.

Jahrling (2001) also suggested that the muscle memory will be forced to correct itself, which leads to long term correction in the foot alignment, posture and control. The SMotO is ideally ‘retraining’ the musculature to maintain the corrected length of muscle. In an article by Best (2005), about interdisciplinary teams getting children back on their feet with the use of surgery, physiotherapy and SMotOs, they noted that the basic philosophy is to let motion happen and to create the possibility to learn and automate correct, physiological motion sequences. This may or may not be possible with children with CP if higher order control inhibits the process.

Clinically, the SMotO have been observed to have improved foot alignment, balance, control with walking and functional skills. Clients were prescribed SMotO as an additional orthosis to AFOs when functional movement was restricted in the AFO. SMotOs have also been prescribed for children who require more feedback from their feet (e.g., hypotonic or hypermobile), where wearing shoes alone has not been effective[3].

There is a lack of evidence examining the ‘sensorimotor response’ paradigm in a paediatric setting, as there are no randomized trials, minimal peer-reviewed papers in English and only a few small cross-sectional paediatric papers (Wegener et al., 2016). There was only one paper in English found that reported the use of SMotOs on in-toeing gait in children (with idiopathic in-toeing or clubfoot). This study found SMotOs improved abnormal gait patterns of paediatric in-toeing gait by decreasing femoral internal rotation through the

[3] Please refer to Chapters 5 & 6 for further background information and the studies relating to SMotOs.

end of the swing phase and the beginning of the stance phase and by decreasing tibial internal rotation during the stance phase (Mabuchi et al., 2012).

The two types of orthoses could not be further apart in design or theory behind effectiveness. On one hand, the AFO can be a bulky orthosis, set to constrict movement around the ankle, (with one plane of movement allowed in designs with a hinge for dorsiflexion), creating the ankle to be a point of stability. Whereas on the other hand, the SMotO has the ability to fit into a typical or orthopaedic supportive shoe, with the design aimed to activate and create movement in the foot musculature. If muscle contributions are not able to overcome the musculature, returning to a more solid shoe may also be used to support idyllic foot position.

1.8. Quality of Life

Increasingly, Quality of Life (QoL) is included as an outcome variable to evaluate the effectiveness of interventions for children with CP (Shelly et al., 2008). QoL could be affected by the reduction in functioning in the aforementioned activity domains, such as physical, intellectual, communication and socialisation.

In a cross-sectional European study (Dickinson et al., 2007), the researchers found there were specific impairments that were associated with poorer QoL in children with CP. These were: children with poorer walking ability had poorer physical wellbeing; those with intellectual impairment had lower moods and emotions and less autonomy; and those with speech difficulty had poorer relationships with their parents. Another study that investigated QoL (Arnaud et al., 2008) found similar domains affected QoL; the severity of motor and intellectual impairment was associated strongly with poor parent reported QoL in the domains of physical well-being, autonomy, and social support, indicating that children with severe impairments are less able to create social time or to maintain relationships with other children.

In support of improving QoL, strength training programmes were found to be effective and beneficial to young people with CP (McBurney, Taylor, Dodd, & Graham, 2003). It was noted that after 6 weeks of exercises (bilateral half squats, heel raises and step ups) the functional benefits included perception of strength, flexibility, posture, walking and the ability to negotiate steps had improved, as well as increase feelings of well-being and improved participation in school and leisure activities. This is supported by Martin, Baker and Harvey (2010) who, through a systematic review, determined that the evidence for strengthening the target muscles groups is stronger, and there is emerging evidence supporting functional training. Therefore, a therapist could conclude that, as per reported domains of QoL, working towards improving the child's strength, functional gross motor skills, gait and communication, will ultimately improve the child's physical ability and QoL.

The literature review above highlights the complexity of CP patients, particularly the paediatric population. While there are many assessment tools available and routinely adopted by physiotherapists and allied health professionals involved in the treatment of paediatric CP patients, there are other assessment tools where there is little evidence in literature to support their use, such as the Anterior Line Method (ALM). Similarly, SMotOs have often been prescribed as a clinically effective orthoses for CP-induced biomechanical alterations but have limited evidence regarding their effectiveness in improving gait and gross motor skills. Taken together, these gaps identified in the literature available warrant further investigation.

2.0. A Proof-of-Concept Experimental Design: Lower Limb Alignment & Orthoses in Paediatric Population

Prelude

The following chapter introduces lower limb alignment and tibial torsion, whereby describing an under researched but frequently used alignment assessment tool. Study 1 continues on to investigate if this alignment technique is reliable and in what population.

2.1. Introduction

Tibial torsion (TT) has been described as the twisting of the tibia about its longitudinal axis (Eyadah & Ivanova, 2011; Li & Leong, 1999; Liu, Kim, Dreup, & Mahadev, 2005; Milner & Soames, 1998; Patel, 2012). Torsion in the tibia is present throughout the lifetime, and a part of normal alignment that changes in the first few years of life (Staheli, Corbett, Wyss, & King, 1985). Torsion through the tibia is one of the factors thought to cause in-toeing (Mabuchi et al., 2012; Son et al., 2014) and out-toeing in children (Sass & Hassan, 2003), whereby the child's foot turns to point excessively medially or laterally, respectively. Throughout the lower limb assessment, the physiotherapist may also uncover other alignment issues such as TT, which may be corrected by orthoses (Mabuchi et al., 2012; Uden & Kumar, 2012). Together, the physiotherapist and podiatrist can assess gait and discuss the best orthosis option for the child, along with a stretching or strengthening regime to complement the effect of the orthoses.

The Anterior Line Method (ALM) is a method that is based on the Talonavicular Method. This technique of lower limb assessment has been used clinically by some therapists and podiatrists to assess underlying factors causing a client's condition, such as TT. The ALM is reportedly used to determine subtalar joint neutral (STJN[4]) and TT through assessing malleolar position (MP) through palpation and visual markings demonstrating alignment (Najjarine & Kielt, 2008), and the basis for orthosis prescription. It needs to be noted that tibial torsion is not confined to a single level. There may be contributions from the pelvis, hip, knee and ankle / foot. To ensure tibial torsion is truly from the tibia, a thorough assessment to rule out other contributing factors will need to be undertaken. The following technique is used once this has been clarified and other torsional contributions eliminated.

Najjarine claims that this approach creates a simple and visually effective method (Figures 5 to 8) of assessing STJN where both the patient and therapist are able to visualise the effect of correction by observing the lines of STJN and when calcaneal alignment is in a resting stance to a corrected 'neutral' stance position. Najjarine and Kielt (2008) also used this technique to assess the MP for diagnosis of TT (both internal and external). TT will be discussed in more detail further in the Appendix C: Tibial Torsion in Children: A Retrospective Study. Therefore, the aim of this chapter was to determine the reliability of the ALM across adults and paediatrics.

[4] For further information on STJN, refer to Chapter 2.2.1.

Figure 5: Resting calcaneal stance position (RCSP) and neutral calcaneal stance position (NCSP) (Najjarine & Kielt, 2008)

A search of the literature revealed that there does not appear to be any studies reporting on the ALM, its reliability or validity as an assessment tool for STJN in the paediatric population. Other than anecdotal evidence, this lack of research restricts the evidence available through which to compare the ALM with other conventional methods.

To ensure assessment and subsequent treatments are worthwhile and progressing towards therapy goals, valid and reliable outcome measures should be used in a paediatric population (Evans, Rome, & Peet, 2012). Validity can be described as the accuracy of the means of measurement, and if that measurement is truly measuring the intended outcome intended (Golafshani, 2003) as well as referring to the strength of the tools' outcomes (Sullivan, 2011). Reliability refers to the replicability or repeatability of results, and to what degree the measurement remains the same, the stability of the measurement over time, and the similarity of the measurements within a time period (Golafshani, 2003).

On this basis, prior to considering the potential use of the ALM to inform this research, the reliability and appropriate use of the ALM to assess STJN needed to be established.

2.2. Lower Limb Assessment: Tools and Techniques

There are a number of different assessment tools available to therapists who work within the paediatric population to assess the lower limb with the assessment tool selected dependant on the presentation or pathology. When assessing the lower limb in a paediatric population, therapists can approach the assessment process as they would an adult with a lower limb musculoskeletal complaint: e.g., observation, lower limb length discrepancy, foot position, muscle control, gait, active range of motion (AROM) and passive range of motion (PROM) in all joints, and structural misalignments (Brukner & Khan, 2001). Commonly used assessment tools in paediatric population that have a specific focus on assessing degrees of spasticity include the Modified Ashworth Scale of Spasticity, Modified Tardieu Scale and PROM (Fosang, Galea, McCoy, Reddihough, & Story, 2003; Yam & Leung, 2006).

Once a therapist has identified possible concerns through their use of dedicated assessment tools (for example, TT), they can progress to additional assessments, treatment and prescription. For results to be considered credible, assessment tools for measurement must be both valid and reliable, with the validity and reliability scientifically examined and reported (Sullivan, 2011). Reliability, defined as the consistency of measurements, can be assessed by the intra-class correlation coefficient (ICC) (Shrout & Fleiss, 1979).

Smith-Oricchio and Harris (1990) note that a reliable measurement tool allows a clinician to make objective and quantifiable recommendations and assists the clinician to assess the intervention progress. Error in assessment may occur when a therapist is using an outcome measure that has not yet been assessed for reliability (or validated), as the measurement may not be accurate or replicable, which could lead to the flow on effect of poor prescription and negative outcomes for the client. For example, a valid and reliable tool that is used in the assessment of lower limb in paediatrics is the Foot Posture Index (FPI-6) (Evans, Rome, & Peet, 2012). The FPI-6 has demonstrated an intra-rater reliability as 'good' (intraclass correlation coefficient [ICC] = 0.93 - 0.94) and an inter-rater reliability as 'largely good' (ICC = 0.79) (Evans, Rome & Peet, 2012), whereas the ALM, another tool reportedly used in the paediatric clinical setting to assess lower limb alignment, does not appear to have any research stating its validity and reliability.

Further details of STJN, and ALM assessment tools, are expanded upon in the following sections to provide context and background to the reliability study of ALM. STJN and its usual assessment tools will be outlined using current literature as a basis. Finally, the ALM will be explained as per its creator's description, with specific focus on providing context for the subsequent reliability study.

2.2.1. Subtalar Joint Neutral

STJN motion has been described in many ways in the literature. Root, Orien, and Weed (1971) outlined STJN as the position where there is neither supination nor pronation in the foot. Similarly, Chen, Yu, Mei, Zhou, and Wang (2008) defined where they define neutral position of a joint as the congruous position between concave and convex surfaces. In contrast, Horner (2000) described the subtalar joint in a more methodical manner as having a total range of motion (ROM) of 30°, 10° of inversion, and 20° eversion and comprises of three articulations between the talus and the calcaneus. STJN and foot position are widely used in the clinical setting as the baseline marker where treatment of lower limb dysfunctions (especially for clinicians involved in orthosis prescription, fabrication and management) require objective and reliable measurements (Sell, Verity, Worrell, Pease, & Wigglesworth, 1994).

Many aspects of clinical assessment of the lower limb are underpinned by positioning and measuring STJN and STJN theory (Harradine, Gates & Bowen, 2018). However, evidence has demonstrated poor (<0.14-0.18) to moderate (0.61- 0.79) intra- and inter-tester reliability for measurements based upon STJN (Harradine and Bevan, 2009; Jarvis, Nester, Jones, Williams & Bowden, 2012; Menz, 1995; Picciano, Rowlands, Worrell, 1993) which is especially relevant to any of the clinical lower limb assessments that rely on bisection lines.

The two main methods of determining STJN are reported as goniometry and palpation (Hunter & Burnett, 2000; Van Gheluwe, Kirby, Roosen, & Phillips, 2002). Van Gheluwe et al. (2002) performed a reliability study using goniometers (legged gravity goniometer, protractor, and a non-legged gravity goniometer) with five raters (of varying years' experience) assessing 30 subjects. This study found poor inter-rater reliability (ICC ≤0.51) for NCSP, but for RCSP the ICC was 0.61 (left) and 0.62 (right). They note a high intra-rater reliability (ICC>0.8) but reported poor measurement accuracy of the clinical measurements commonly used, which led the authors to question the practical usefulness and the validity of these clinical measurements.

Palpation is the most common method used to locate the neutral position of the subtalar joint (Hunter & Burnett, 2000; Pierrynowski & Smith, 1997), which has low to moderate reliability (ICC ranged from 0.25 to 0.60) in prone (Smith-Oricchio & Harris, 1990). Other factors that have been reported to influence the measurements of STJN include joint ROM, false talar head, the presence of a forefoot valgus or varus (Hunter & Burnett, 2000) and if the assessment was in prone or weight-bearing (Smith-Oricchio & Harris, 1990). Menz (1995) notes that there are several factors influencing accuracy of assessment such as skin movement, pen marker thickness and practitioner dexterity. In addition to these factors, it has also been reported that assessing the paediatric population can affect measurement (Morrison & Ferrari, 2009), especially if there is low experience in assessing and treating paediatric populations.

Sell et al. (1994) agreed that foot and ankle evaluation relied on a proper assessment of the subtalar joint. The authors performed a reliability test between two techniques (weight-bearing navicular height and calcaneal position) with an inclinometer on 30 volunteers (n=60) and discovered that their inter-rater reliability for calcaneal position ranged from 0.68 – 0.91 (95% Confidence Interval [CI]), and navicular height 0.73 – 0.96 (95% CI), concluding that the weight-bearing measurement techniques are reliable and acceptable.

2.3. Inter- and Intra-Rater Reliability of the Anterior Line Method

2.3.1. Introduction

STJN may be a position used by clinicians to align a patient's foot to obtain a foot cast prior to fabrication of orthosis (Chen et al., 2008; Hunter & Burnett, 2000) and to obtain relevant lower limb measures (Hunter & Burnett, 2000). Considering this, there is no actual consensus on what STJN actually is with the position described in many ways in the literature. While Root, Orien, and Weed (1971) and Chen et al. (2008) employed similar definitions, the definition by Horner (2000) differs as STJN was noted to be 20° inversion and 10° eversion. Considering this, a lower limb assessment of STJN within the clinic setting generally relies upon talar head palpation method or talonavicular palpation method.

The ALM includes both a physical and visual assessment, which clinicians claim can create more accuracy in determining STJN when compared to palpation alone, as it will not be confounded by the presence of a false talar head (Najjarine & Kielt, 2008, p. 24). Other lower limb assessments, such as the FPI-6 technique (although not used to determine STJN, is a valid and reliable tool to assess the foot posture), relies upon palpation and observation of anatomical markings, whereas the reference points applied to the dorsal surface during the ALM provides a visual reference point during assessment.

However, with the aforementioned lack of any known measures of reliability, the need to ensure that the ALM can be measured reliably is paramount if it is to be used as an assessment tool to determine STJN. In addition, a determination of the reliability of this tool can be used to inform future validity studies.

It was hypothesised that the ALM would be more reliable when implemented by more experienced assessors. As most of the assessors' experience of the ALM, clinically, has been in the adult population, adults will be included in this study as a comparison point. On this basis, the aim of this study was to assess the intra- and inter-tester reliability when using the ALM (an anecdotal, non-researched method) through using a proof-of-concept experimental design on both adults and children in order to determine if the ALM has practical potential in this population.

2.3.2. Methods

2.3.2.1. *Design*

Ethics approval was sought and approved through Bond University Human Research Ethics Committee (RO-1539). This proof-of-concept experimental design consisted of six assessors from four professions who performed a test / re-test on the left and right feet of 21 subjects (6 children and 15 adults).

2.3.2.2. *Recruitment*

A number of strategies were used to recruit subjects for data collection. Letters were sent to several allied health professionals who were known to use the ALM to draw their attention to the study. This letter offered information regarding the purpose of the study, and an offer to participate in assessing the participants. An information poster was displayed at the location of assessment which invited readers to inquire at reception about the study and for further information. All participants were required to have given informed consent prior to participating in the study and were given an explanatory statement and consent form to complete prior to data collection.

2.3.2.3. *Participants*

The inclusion criterion for this study was a healthy foot and ankle absent from any musculoskeletal injury. The exclusion criteria included: a) A history of triple arthrodesis, b) forefoot amputation, or any history of surgery that may alter the tibial crest or 2nd metatarsal, c) arthritic or hypo-mobile ankle joint, d) if the assessor was unable to locate the 2nd metatarsal, and e) the subject had a high infection risk, or skin allergy to testing material (whiteboard marker). It is important to note that supination or pronation was not an exclusion criterion and all patients with a supinated or pronated foot were included in the study.

2.3.2.4. *Assessors*

Of the six assessors, there were three podiatrists, one physiotherapist, one osteopath and one chiropractor, all of whom had undertaken the training course for ALM and utilised the ALM in their clinic for a minimum of one year. Assessors 1, 3 and 4 were podiatrists (A1, A3, A4), Assessor 2 (A2) was a physiotherapist, Assessor 5 (A5) was an osteopath, Assessor 6 (A6) was a chiropractor. A1 had the most experience with using the ALM (20 years) and used it regularly on both and adult and paediatric population, whereas the other podiatrists, A3 and A4, had two- and six-years' experience respectively. A2 worked with children as a physiotherapist and had been using the ALM for three years. A5 and A6 had both used the ALM sporadically in their practice over the past year.

Due to the study design, there was no chance of contamination of results and potential manipulation of results. All data were collected, collated and analysed by the researcher.

2.3.2.5. *Protocol*

The lead researcher screened each participant prior to data collection to ensure eligibility against the inclusion criterion and exclusion criteria. Each subject was allocated a private room and number for randomisation. The subjects were randomly allocated to a number and rotated through the assessors by a randomized allocation system generated by a third party. The assessors then had an increased chance of 'forgetting' the previous measurements that had been taken on the first round of assessments.

Blinding was used to reduce bias between the assessors. This was ensured by using removable markers that were wiped with an 'Alco-wipe' after each assessment has been performed by the assessor prior to the next allied health professional taking measurements.

2.3.2.6. *Outcome Measure*

The ALM was carried out twice by the six assessors, providing the values for the Resting Calcaneal Stance Position (RCSP) and the Neutral Calcaneal Stance Position (NCSP). The ALM was measured using the protocol described below with all bony landmarks located by palpation and marked by pen:

1) Bony landmarks located by palpation are marked by a pen on the skin
    a. The depression on lateral side of talus head and anterior to malleolus (Figure 6).

    The depression on medial side of the talus head and anterior to the medial malleolar (Figure 6).

Figure 6: Marked head of talus (blue) and presence of false talar head (red) and marked alignment of the calcaneus (Najjarine, Finding NCSP Using the NAS Anterior Lines Method)

2) The centre of these two points is marked, giving the centre of the ankle.

3) The 2nd metatarsal head (dorsal surface) is joined to the centre point at the bisection of the talonavicular reference points by a dotted line.
4) The apex of the anterior tibial crest (only concentrating on the lower one third of the tibia) is joined to the centre point of the ankle via a dotted line.
5) The bisection of the calcaneus aligned and marked (Figure 7).

Figure 7: Bisection of the calcaneus and dorsal aspect of foot (resting calcaneal stance position) (Najjarine & Kielt, 2008)

When the lines on the anterior shin and dorsal aspect of the foot are matched to create a straight line, the rearfoot angle and bisection of the lower 1/3 and calcaneus align (Figure 8). The calcaneal line is measured with a protractor giving an angle measurement of NCSP.

Figure 8: Neutral calcaneal stance position (Najjarine & Kielt, 2008)

Subjects were asked to stand normally and the RCSP was measured using a protractor at the line of bisection of calcaneus and recorded (Figure 9).

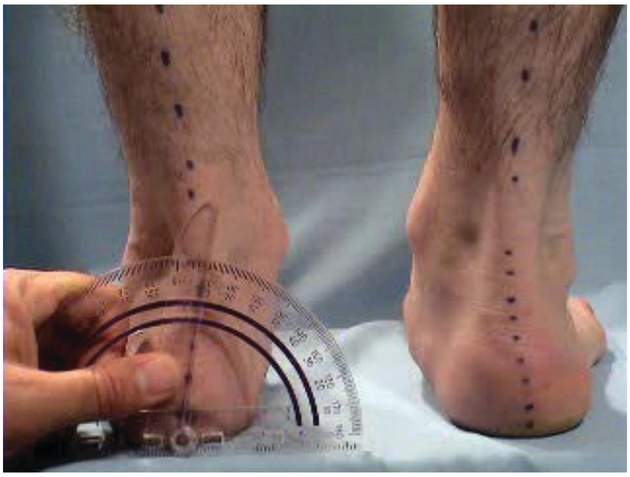


Figure 9: NCSP with protractor measurement (image courtesy of Rod Kielt, 2021)

Assessors requested subjects to assist alignment by moving their ankle to create a straight line from the tibia and dorsal surface of the foot. Once the assessor was happy with the alignment, they measured the NCSP by using a protractor at the bisection of calcaneus line.

Each assessor took two measurements per foot of the subject. The recording sheets were collected by the lead researcher and each assessor was given a new sheet for each re-measure, therefore ensuring the assessor was blinded to previous assessment measurements taken by the other assessors, as well as reducing the possibility of remembering their own previous measurements. RCSP and NCSP were recorded, and intra- and inter-rater reliability was assessed statistically using software program Statistical Package for Social Sciences (SPSS), using one-way analysis of variance (ANOVA), an ICC estimate based on mean squares (3,1), and the standard error of measurement ($p<0.05$). According to Munro (2004), the strength of an agreement is very low if the correlation ranged from 0 to 0.29, a low correlation if 0.30-0.49, a moderate correlation if 0.50-0.69, a high correlation if 0.70-0.89 and a very high correlation if 0.90-1.00.

2.3.3. Results

2.3.3.1. *Participant Demographics*

Of the 21 participants, there were 15 adults (mean = 39.2 ± 13.87 years; range 24 years to 65 years,) and six children (mean = 11.67±2.25 years; range 8 years to 14 years), meaning a total of n=42 feet (Table 3).

Table 3: Demographics of ALM participants (mean ± SD, n = 21)

Adult / Child	n =	Age (years)	M/F
Adult	15	39.2 ± 13.87	3 M 12 F
Child	6	11.67 ± 2.25	2 M 4 F

M=male; F=female; SD=standard deviation

The intra-rater reliability for RCSP for all subjects (Table 4) ranged from moderate (0.51; 95% CI: 0.11 to 0.77) to very high (0.96; 95% CI: 0.93 to 0.98) across all six Assessors, and a low correlation (0.30; 95% CI: 0.12 to 0.64) to high correlation (0.86; 95% CI: 0.75 to 0.92) for the NCSP across Assessors. A6 demonstrated moderate intra-rater reliability for RCSP (0.51; 95% CI: 0.11 to 0.77) and showed poor intra-rater reliability for NCSP (0.30; 95% CI: 0.54 to 0.84) (Table 4).

Table 4: Intra-rater reliability RCSP (°) and NCSP (°)

Assessor	RCSP (ICC; 95%CI: range)	NCSP (ICC; 95%CI: range)
1	0.96; 0.93-0.98	0.86; 0.75-0.92
2	0.85; 0.74-0.92	0.55; 0.30-0.73
3	0.81; 0.64-0.90	0.59; 0.36-0.76
4	0.65; 0.44-0.79	0.72; 0.54-0.84
5	0.73; 0.33-0.91	0.65; 0.19-0.83
6	0.51; 0.14-0.77	0.30; -0.12-0.64

RCSP=Resting Calcaneal Stance Position; NCSP=Neutral Calcaneal Stance Position; ICC=Intraclass Correlation; CI=Confidence Interval; °=degrees

A6 was ineligible for further in-depth assessment of the ALM when assessing the paediatric population due to their inexperience and lower reliability to ensure that this inexperience was not a factor for the ALM to be deemed unreliable. A5 was unable to attend the in-depth paediatric data collection session. Therefore, only A1, A2, A3 and A4 were compared (Table 5).

Table 5: Average RCSP and NCSP in paediatric population

Assessor	RCSP (avg°)	NCSP (avg°)
1	-3.33	3.75
2	-2.96	3.42
3	-4.08	2.25
4	-4.13	0.42

RCSP=Resting Calcaneal Stance Position; NCSP=Neutral Calcaneal Stance Position; avg=average; °=degrees; -ve = pronation, +ve = supination

Assessing the average of the two measures, the intra-rater agreement varied amongst assessors (p=0.011) in the adult population. Post Hoc analyses identified very high agreement between three of the assessors (A1, A2 and A3), and low agreement for NCSP (p=0.001) when A1 to A3 were compared with A4, which may be due to a data recording error as the raw numbers were markedly different (Table 5). The average RCSP when measured in children (one-way ANOVA) found to be insignificant (p=0.485), between A1 through A4.

2.3.4. Discussion

The hypothesis that the ALM would be more reliable when implemented by more experienced assessors was confirmed. The results demonstrate that the RCSP appears to be more reliable to measure having a high mean overall result of 0.75 compared to a moderate mean overall results of 0.61 for NCSP across the ages. These results are surprising given there have been numerous investigations reporting poor reliability of NCSP and RCSP using bisection of the calcaneus line due to issues from pen thickness, skin movement to the frontal plane angle of the calcaneus is not true to the calcaneal bisection (Menz, 1995; Picciano, et al., 1993; Jarvis, 2013).

The ALM uses both palpation and visual marking with a protractor, similar to the two main methods of determining STJN reported previously (Hunter & Burnett, 2000; Van Gheluwe et al., 2002). Sell et al. (1994) performed a reliability study on two methods to assess STJN – navicular height and calcaneal position with an inclinometer and found that weightbearing measurements were reliable for clinic and research purposes. Using the ALM as a weightbearing measurement (as per protocol) may have contributed to the strong reliability of the two measurements and may only be indicated in a weight bearing environment. The ALM study used static positions to assess STJN and, as such, is only one plane of movement. Considering this joint is triplanar and used in dynamic movements, this presents a problem when comparing static measurements to dynamic pathologies (Jarvis et al., 2017) and may not be clinically relevant. Jarvis et al. (2017) challenged the validity of the relationship between positional foot deformities (such as inverted subtalar joint in NCSP) and the foot kinematics during gait (such as rearfoot kinematics during midstance). They argue that the differences in forces applied to the foot in static versus forces and muscle contractions during dynamic movements bear little resemblance to each other. Jarvis et al. (2017) found that only 39% of feet passed through STJN during stance when assessed using three-dimensional gait analysis. Future studies could investigate the use of ALM in a more dynamic setting.

The assessors report that this was an easy method of alignment to understand. However, there were limitations to its implementation. These included assessors potentially remembering previous results. Further throughout the data collection, the subjects reported that relaxing into RCSP was feeling harder to accomplish and recognise, as they had aligned their feet and ankles continuously. When assessors were collecting data from the children, they found that following the direction for alignment was attempted by all children, but the younger participants needed assistance to hold the position for measurement collection.

The results show that the RCSP appears to be more reliable to measure than the NCSP. This may be due to factors such as the subjects being able to hold a resting position more easily than the corrected position or the assessors may not have aligned the landmarks correctly. Therefore, when the subject attempts to align markings, the NCSP could be subject to error. Another contributing factor to the increased reliability in the dynamic measurement could be the assessors professional handling experience, and therefore, ability to manipulate or direct the neutral alignment. The ALM's reliability may also be limited by structural deformities or presence of other malalignment which could affect the visual and physical alignment for the ALM.

The results indicate that the level of experience in using the ALM and the profession of an assessor, influences the reliability of measure. The highest reliability was A1 who had the most experience, followed by a younger podiatrist who practices the technique multiple times a day. The poorest reliability being from the chiropractor who has attended a single workshop and only occasionally uses the technique in his practice. Between the physiotherapist and podiatrists, the reliability was high for all measurements. This may be due to both professions having extended experience with children and lower limb assessment. Due to the variability of individual results, it may be suggested that the feasibility of this method might be only suitable for clinicians with proper training and experience.

Comparing the ALM against the FPI-6, it is noted that the FPI-6 uses a visual assessment of the lateral anatomical line of the ankle to determine pronation, whereas the ALM uses a visual cue from a line that has been drawn on anatomical landmarks, which may allow for error in the inexperienced assessor. Both have the element of visual alignment, but the ALM relies upon the subject being able to follow direction, balance in standing, and control the movement of their foot to create alignment. This assumption creates difficulty in reliability especially when applying this measurement to those with any motor or intellectual deficits that could affect the accuracy of instruction and response. Having a structured training protocol and recommended practice timeline would assist in eliminating the factor of inexperience in the quality of measurements.

Finally, this study's results showed that the ALM is more reliable when measuring adult subjects when compared to children across the six assessors of varying experience. Adults were included in this study as they were the population this technique was commonly used on in the clinical setting. They were also included to see if there was a difference in the reliability between adult and paediatric populations. Specifically, with the ALM being assessed by those less experienced, improved reliability may be due to adults being able to follow and co-ordinate directions better, or larger landmarks allow for easier palpation and thus creating more accurate alignment.

The lower paediatric measurement reliability aligns with Morrison and Ferrari (2009) who noted that assessing a paediatric population affects reliability of measurements. Lower reliability was noted in the ALM when the assessors had less experience. Although, the most experience assessors were found to have a higher reliability regardless of subject, demonstrating that experience in the ALM and assessing children is a key factor in the reliability of this technique. A need to research the ALM technique with a larger recruitment of children appears warranted. In particular, as all participants were healthy, further investigation within different populations, such as CP who may have reduced intellect, reduced body control and abnormal foot positioning, would greatly enhance the viability of this method to be used across a greater population and varying clinical settings.

2.3.4.1. Limitations

A limitation of this study was the small number of children participants, especially given the focus of this thesis. This was due to time constraints and a reduced number of those who consented to participate. Another notable limitation of this study was that the validity of the ALM still needs to be established. Future research could include replication of the study using standing x-rays, which could serve as gold standard to determine validity. To then assess the reliability, a standardised photo, such as through a phone app could be used to compare the RCSP and NCSP measurements to the gold standard. This may then improve the overall reliability and improve the accuracy of ankle angle measurements and therefore provide a more reliable result. Due to the STJ being dynamic, and as the ALM is a static measure, the ALM would need to be validated for use in a dynamic setting.

2.3.5. Conclusion

The ALM was found to have poor reliability in a paediatric population, especially when used by an inexperienced assessor. Therefore, the findings of this study suggest that the use of the ALM in the assessment of children may not be recommended unless there is extensive experience by the assessor in both the ALM technique and in assessing the paediatric population. The level of experience and familiarity of using the ALM may influence reliability, as well as when measuring adult subjects compared to children.

To assess across different populations, such as children with CP, the ALM would require an alternate version or modification as the participant requirements, expected response and control of lower limb may be affected, thus affecting the reliability and specificity of the ALM. Furthermore, this tool should be used with caution in children with CP due to factors that impact its reliability: factors such as poor comprehension of instructions, balance requirement in standing, inability of patient to consciously override muscles to create fine-tuned alignment, and CP related spasticity.

Due to the limited reliability and the extra requirements needed to assess children with CP, the feasibility of using the ALM in this specific population was not ideal. As such, the ALM was considered unsuitable as an outcome measure in the subsequent research and was not implemented in further studies as originally planned, when assessing children with CP. Further research would be required to investigate feasibility and reliability of this technique in a typical paediatric population, as well as in specialised population such as children with CP.

3.0. Orthoses, Gait & Children with Cerebral Palsy: A Narrative Systematic Review

Prelude

The following chapter presents an investigation and appraisal of the literature surrounding lower limb orthoses in children with cerebral palsy through a narrative systematic review. The initial aim of this review was to identify and report on studies investigating the use of sensomotoric orthotics and their subsequent effects on gait and gross motor skills in children with cerebral palsy. However, the initial search was unable to identify research in this specific field. As such, the aim of the review was modified to a) identify the types of lower limb orthoses typically used, and b) determine the effect of identified lower limb orthoses on gait and gross motor skills in children with cerebral palsy.

3.1. Abstract

Lower extremity orthoses are often used to address the biomechanical limitations children have due to cerebral palsy. What are the various types of lower limb orthoses used to improve gait and gross motor skills? A search of eight online databases was performed between May and June 2019. Included studies were critically appraised. These studies were also classified for their level of evidence using the Oxford Centre for Evidence-Based Medicine Levels of Evidence classification system. Eight studies were assessed for methodological quality. Seven studies of good quality (67%-85%) were included in the qualitative synthesis. The levels of evidence ranged from Level 1b (Randomised Control Trial design) to Level 4 (quasi-experimental designs), with ranges of 15 – 38 participants and aged 21 months to 15 years. Overall, all orthoses demonstrated some level of improvement but orthoses using a more dynamic approach were found to be effective in improving gait and gross motor skills. Orthoses intervention using were found to be effective in improving gait and gross motor skills in children with cerebral palsy. There is a potential benefit to the clearer defining of ankle-foot orthoses prescription within research studies.

3.2. Introduction

Cerebral palsy (CP) is a term used to describe a group of permanent, non-progressive, disorders affecting the development of human movement and postures, caused early in life primarily by a brain injury or lesion (Danino et al., 2015). CP leads to physical activity limitations (Christovao et al., 2015; Ridgewell, Dobson, Bach, & Baker, 2010; Romkes & Brunner, 2002; Rosenbaum et al., 2007; Wingstrand, Hägglund, & Rodby-Bousquet, 2014) through spasticity, muscle weakness, impaired postural control, and selective motor control (Wingstrand et al., 2014) and a common treatment is the use of lower limb orthoses (LLO) (Morris, 2002).

The most common LLO is the ankle-foot orthoses (AFO) (Ries, Novacheck, & Schwartz, 2015). Common styles of AFOs (Wren et al., 2015), include solid-AFOs (SAFOs), hinged-AFOs (HAFOs) with dorsiflexion or plantarflexion stops, Posterior Leaf Spring (PLS) and dynamic-AFOs (DAFOs). The aims of AFO management in children with CP are to; correct and/or prevent deformity (joint alignment), provide a base of support, facilitate training of motor skills, and improve efficiency of walking (Bjornson et al., 2016; Bjornson, Schmale, Adamczyk-Foster, & McLaughlin, 2006; Brodke et al., 1989; Buckon, Jakobson-Huston, Moor, Sussman, & Aiona, 2004; Figueiredo, Ferreira, Moreira, Kirkwood, & Fetters, 2008; Knutson & Clark, 1991; Morris & Condie, 2009; Rethlefsen, Kay, Dennis, Forstein, & Tolo, 1999; Ries, Novacheck, & Schwartz, 2015; Romkes & Brunner, 2002; Smith et al., 2009; Westberry et al., 2007; White, Jenkins, Neace, Tylkowski, & Walker, 2002; Wingstrand et al., 2014). AFOs also enable a continuous Achilles / gastrocnemius stretch (Boyd, Pliatsios, Starr, Wolfe, & Graham, 2000; Hainsworth, Harrison, Sheldon, & Roussounis, 2007; Morris, 2002), position the foot for function (Brodke et al., 1989; Kane, Musselman, Manns, & Lanovaz, 2016; Knutson & Clark, 1991; White et al., 2002), and prevent pain (Westberry et al., 2007). AFOs comprise a polypropylene cast that covers the entire posterior and mediolateral borders of the calf, sole of the foot, with straps across the anterior upper tibia and front of the ankle (Radtka, Skinner, Dixon, & Johanson, 1997).

Another LLO is the Postural Control Insole (PCI). PCIs were investigated by Christovao et al. (2015) in a randomized controlled trial. The researchers found PCIs led to an improvement in static balance and performance of the Timed Up-and-Go (TUG) in children with CP. Outcome measures are used to assess gross motor skills (GMS), such as the Gross Motor Function Measure (GMFM), Pediatric Evaluation of Disability Inventory (PEDI), Pediatric Outcomes Data Collection Instrument (PODCI), TUG, temporal-spatial gait parameters and O_2 cost of walking (Alotaibi, Toby, Kennedy, & Bavishi, 2014; Dhote, Khatri, & Ganvir, 2012; Oeffinger et al., 2004, 2007, 2008).

However, there is no consensus as to which is the most effective type of LLO to improve both gait and GMS in children with CP. It was hypothesised that lower limb orthoses would improve alignment, cadence and balance. Therefore, this review aimed to identify and critically appraise studies investigating the use of LLOs in both gait and GMS in children with CP and synthesise their findings.

3.3. Methods

The approach taken for this systematic review was based on the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) statement (Moher, 2009), outlining a high-quality ordering of literature searches. The methods and eligibility criteria for included studies were detailed in advance in a protocol registered at The Centre of Reviews and Dissemination (CRD) at the University of York – PROSPERO (CRD42019137970) (CRD, 2009).

The inclusion criteria for the purpose of this review were kept broad to minimise selection bias. Table 6 describes the eligibility criteria. The outcome measures selected were deemed by researchers to be the most clinically applicable, as well as being those validated for this population (Ko & Kim, 2013). The results of the search, screening and selection processes are documented using the PRISMA flow diagram (Figure 10) (Moher, 2009).

Table 6: Eligibility criteria

Inclusion Criteria	Exclusion Criteria
Study reported original research	Study included the use of botulinum toxin therapy
Study was available in full text and in English (or translatable to English by a service)	Treadmill suspension system used
The study was a trial	Garment orthotic (not lower limb) used
The study included children with CP between ages 0-18 years with any type or level of function and were using lower limb orthoses	Study designs other than trials
The studies investigated at least one type of LLO, and compared to barefoot, a control or other LLO	Only 1 of gait kinematic or kinetic data or outcome measures only (not relating to gross motor function, or not comparing orthoses)
Reported at least one of the following outcome measures: GMFM / visual gait assessments / functional ability and gait parameters	Paper was published 20 years ago or more (0-1999)

CP = Cerebral Palsy; LLO = lower limb orthoses; GMFM = Gross Motor Function Measure

Key search terms were entered into selected databases (CINAHL, SPORTDiscus, EMBASE, Web of Science, Scopus, ProQuest, PubMed and Cochrane Library) between May and June 2019 in order to capture research relevant to this review (see Table 7 for further information on search terms). The key search terms used, alone or in combination, were derived from previously known literature on this topic and from discussion with subject matter experts in this field. The parameters of the systematic review were designed to accommodate LLO with a particular focus on quality of gait and assessments used to evaluate GMS changes.

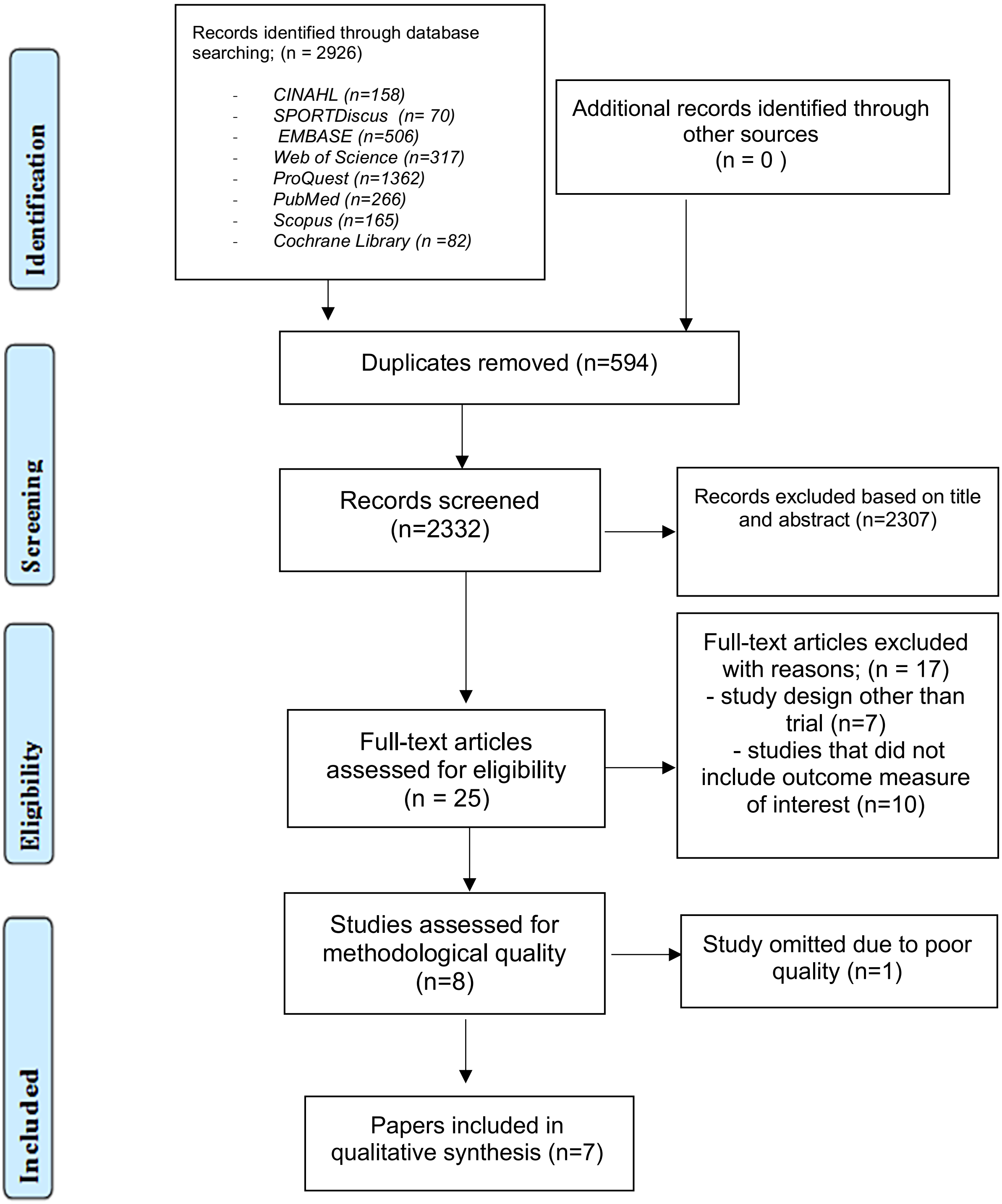


Figure 10: PRISMA diagram (Moher, 2009) detailing results of the search, screening and selection processes

3.3.1. Data Extraction and Methodological Quality Analysis

The main author extracted all relevant information into a spreadsheet; type and level of CP, age of participants (any variables such as mobility aids etc), types of orthoses, comparator (control, placebo, other orthoses), types of outcome measures and results, and study design.

The modified Downs and Black (Downs & Black, 1998) checklist was used to assess the methodological quality of the included. This methodological protocol employs a 27-question checklist to assess 5 key areas of methodological quality; statistical power, internal validity (bias and confounding), external validity and reporting quality. For the purposes of this study, the scoring for Question 27 (reporting a power analysis) was modified to ‘0’ points for a ‘no’, or ‘1’ point if the authors did report their power analysis. This modified approach to the checklist has been used previously to limit subjectivity in scoring (Lyons, Radburn, Orr, & Pope, 2017). As such, the maximum possible raw score became 27, as opposed to the original maximum score of 32.

The final score was then graded based upon the approach described by Kennelly (2011). Kennelly’s (2011) grades were categorized as ‘poor’, ‘fair’ and ‘good’, with the methodological quality rating determined by the scores assigned in the Downs and Black protocol. However, as the maximum score for this review was modified to 27 points, the Kennelly grades were modified and converted to percentages to accommodate the modified Downs and Black checklist total raw score. On this basis, the grading system applied was as follows: >61% was graded as ‘good’ quality, 45-61% as ‘fair’ quality, and <45% as ‘poor’ quality. This modification has been previously used in critical reviews (Lyons, et al., 2017) and formed the basis of the critical appraisal score (CAS) for this review.

All studies were independently rated by 2 authors independently (CM, VS) using the Downs and Black protocol (Downs & Black, 1998) with the level of agreement between raters measured using a Kappa analysis of all raw scores (27 scores per paper) by the third author (RO). If there was a failure to reach consensus, final scores were moderated by the third author (RO). As the hierarchical system of classifying evidence is paramount to evidence-based medicine, the studies were also subjected to a ‘level of evidence’ grading using the Oxford Centre for Evidence-Based Medicine Levels of Evidence (EBMLE) classification system (Howick et al., 2011).

Table 7: Details of the database search terms

DATABASES	SEARCH TERMS
CINAHL **SPORTDISCUS**	("cerebral palsy" OR (MH "Cerebral Palsy+")) AND ((MH "Pediatrics+") OR pediatric* OR paediatric* OR child OR children OR (MH "Child+")) AND (Sensorimotor* OR sensomotor* OR orthosis OR orthoses OR orthotic* OR brace* OR insole* OR (MH "Orthotic Devices+")) AND (foot OR feet OR ankle* OR (MH "Foot+") OR (MH "Ankle Joint+"))
EMBASE	("cerebral palsy" OR 'Cerebral Palsy'/exp) AND ('Pediatrics'/exp OR pediatric* OR paediatric* OR child OR children OR 'Child'/exp) AND (Sensorimotor* OR sensomotor* OR orthosis OR orthoses OR orthotic* OR brace* OR insole* OR 'Orthotic Devices'/exp) AND (foot OR feet OR ankle* OR 'Foot'/exp OR 'Ankle Joint'/exp)
WEB OF SCIENCE	("cerebral palsy" OR "Cerebral Palsy") AND (Pediatrics OR pediatric* OR paediatric* OR child OR children OR Child) AND (Sensorimotor* OR sensomotor* OR orthosis OR orthoses OR orthotic* OR brace* OR insole* OR "Orthotic Devices") AND (foot OR feet OR ankle* OR Foot OR "Ankle Joint")
PROQUEST	("cerebral palsy") AND ("pediatric" OR "pediatrics" OR "paediatric" OR "paediatrics" OR "child" OR "children") AND ("orthosis" OR "orthoses" OR "orthotic" OR "orthotics" OR "brace" OR "braces" OR "insole" OR "insoles" OR ("sensorimotor*" AND "orthos*s") OR ("sensorimotor*" AND "orthic*")) AND ("foot" OR "feet" OR "ankle*") AND ("gait") AND ("gross motor")
PUBMED	("cerebral palsy" OR "Cerebral Palsy"[Mesh]) AND (Pediatrics[Mesh] OR pediatric* OR paediatric* OR child OR children OR Child[Mesh]) AND (Sensorimotor* OR sensomotor* OR orthosis OR orthoses OR orthotic* OR brace* OR insole* OR "Orthotic Devices"[Mesh]) AND (foot OR feet OR ankle* OR Foot[Mesh] OR "Ankle Joint"[Mesh])
SCOPUS	("cerebral palsy") AND ("pediatric" OR "pediatrics" OR "paediatric" OR "paediatrics" OR "child" OR "children") AND ("orthosis" OR "orthoses" OR "orthotic" OR "orthotics" OR "brace" OR "braces" OR "insole" OR "insoles" OR ("sensorimotor*" AND "orthos*s") OR ("sensorimotor*" AND "orthic*")) AND ("foot" OR "feet" OR "ankle*") AND ("gait") AND ("gross motor") AND NOT INDEX(medline)
COCHRANE	("cerebral palsy" OR [mh "Cerebral Palsy"]) AND ([mh Pediatrics] OR pediatric* OR paediatric* OR child OR children OR [mh Child]) AND (Sensorimotor* OR sensomotor* OR orthosis OR orthoses OR orthotic* OR brace* OR insole* OR [mh "Orthotic Devices"]) AND (foot OR feet OR ankle* OR [mh Foot] OR [mh "Ankle Joint"])

3.4. Results

Eight studies (see Figure 10) met the eligibility criteria and were assessed for methodological quality through the modified Downs and Black (Downs & Black, 1998) checklist and graded according to Kennelly (Kennelly, 2011). One paper (Russell & Gorter, 2005) was removed after applying the modified Downs and Black checklist due to a less than acceptable rating (13/27), leaving papers for final inclusion.

The seven studies included three randomized controlled trials (Buckon et al., 2001, 2004; Christovao et al., 2015), 2 quasi-experimental studies with pre-/post- design (Dalvand, Dehghan, Feizi, Hosseini, & Armirsalari, 2013; Smith et al., 2009;), and 2 randomised crossover trials (Bjornson, Schmale, Adamczyk-Foster, & McLaughlin, 2006; Zhang, Wang, Yang, & Zhao, 2009). An overview of the characteristics of each paper can be seen in Table 8. How each study performed across the 5 key areas of the modified Downs and Black (Downs & Black, 1998) appraisal tool, final raw scores, final CAS based on the modified Kennelly grading system (Kennelly, 2011), and EMBLE results are detailed in Table 9. The 2 reviewers score of the papers was assessed for consistency. Cohen's k was run to determine the level of agreement between the 2 reviewers. A tabulated summary of each study's statistical analysis, results, strengths and limitations can be found in Table 10. There was a 'strong' (k=0.903, p<0.0005) level of agreement (Viera & Garrett, 2005) found between the 2 reviewing authors (CM, VS) scores.

3.4.1. Demographics of the reviewed studies

The final yield of papers (n=7) included four studies from the U.S.A. (Bjornson et al., 2006; Buckon et al., 2001, 2004; Smith et al., 2009), and one each from Iran (Dalvand et al., 2013), Brazil (Christovao et al., 2015) and China (Zhang et al., 2009).

Participants included in these studies were from the public health sector (e.g. hospitals) with all seven reporting on children with spastic CP; five (Buckon et al., 2004 Christovao et al., 2015; Smith et al., 2009; Zhang et al., 2009) out of seven studies reported on diplegia alone, and one (Buckon et al., 2001) on hemiplegia and one (Bjornson et al., 2006) reported on mixed types of CP, with the children in the seven studies ranging in age from 1.9 years (Bjornson et al., 2006) to 15 (Buckon et al., 2001) years of age. Two studies (Bjornson et al., 2006; Dalvand et al., 2013) included children with Gross Motor Function Classification System (GMFCS) levels I to III, whereas there were three studies (Buckon et al., 2004; Christovao et al., 2015; Zhang et al., 2009) that only included children with GMFCS

I and II, and 1 that only included GMFCS I (Smith et al., 2009). There was only one study (Buckon et al., 2001) that did not disclose levels of GMFCS.

There were two studies (Dalvand et al., 2013; Zhang et al., 2009) that implemented therapy. One (Dalvand et al., 2013) used occupational therapy (36 training sessions) as an additional intervention along with orthoses and the other (Zhang et al., 2009) utilised strength rehabilitation training. Only two studies (Christovao et al., 2015; Smith et al., 2009) used a control group. Smith et al. (2009) had neurotypical children as the control group and Christovao et al. (2015) used the same population as the intervention group.

The size of the study groups ranged from 15 (Smith et al., 2009) to 38 (Zhang et al., 2009) participants and the length of the studies from one day (Bjornson et al., 2006) to 10 weeks (Smith et al., 2009) and three (Buckon et al., 2001; Dalvand et al., 2013; Zhang et al., 2009), four (Christovao et al., 2015) and 12 months (Buckon et al., 2004).

3.4.2. Orthotic Types used in the reviewed studies

Along with the length of orthotic use, the type of orthotics used differed between studies. DAFO were used in the study by Bjornson et al. (2006) whereas Smith et al. (2009) compared DAFO and HAFO. Dalvand et al. (2013) and Zhang et al. (2009) used HAFO and SAFO, whereas Buckon et al. (2001, 2004) used HAFO, SAFO and PLS in their comparative studies. PCIs were used by Christovao et al. (2015).

3.4.3. GMFM

All studies included used varying combinations of functional skill outcome measure and gait assessment. All studies included elements of the GMFM in their outcome measures. However, only one study (Christovao et al., 2015) utilized the GMFM-88 as their main outcome measure and did not find any significant changes in scores between intervention and placebo insole. The remaining six studies used only sections "D" and "E" of GMFM-88 to assess standing and walking.

Three studies (Bjornson et al., 2006; Dalvand et al., 2013; Zhang et al., 2009) investigating DAFOs or HAFOs compared to SAFOs found significant changes in the GMFM. Bjornson et al. (2006) also used the GMFM-66 and notes percentage scores for all dimensions were significantly higher when the patients were wearing the DAFOS (p>0.001) compared to SAFOs. Zhang et al. (2009) found GMFM scores in HAFOs group patients were significantly better than those in SAFOs group after three months of treatment (p<0.01). Zhang et al. (2009) found that after wearing the ankle-foot orthosis, the scores of the two functional areas of the GMFM in the HAFOs group were significantly higher than those in the SAFO group ($p<0.05$). The study by Dalvand et al. (2013) also found a significant difference when comparing HAFOs group with SAFOs ($p<0.05$) and the control groups ($p<0.01$) in improvement of gross motor function, but there was no significant difference between SAFOs group and the control group ($p=0.631$).

Opposing this, Buckon et al. (2001) found gross motor function skills were not significantly altered in any of the AFO configurations and Buckon et al. (2004) noted that AFO use did not significantly improve skills within the standing dimension of the GMFM. Buckon et al. (2001) reported similar results as Buckon et al. (2004), that all AFO configurations significantly improved performance of the motor skills within the walking / running / jumping dimension of the GMFM compared with the barefoot condition ($p<0.005$) (Buckon et al., 2001), ($p<0.002$) (Buckon et al., 2004).

Contrary to this, Smith et al. (2009) evaluated sections D&E and found that regardless of treatment, the children exhibited high GMFM scores, with no significant differences noted between HAFO and DAFO.

3.4.4. Visual Gait Assessment

There were no studies reporting on visual gait assessment to compare orthoses.

3.4.5. Functional Ability

Other functional outcome measures included PEDI (Buckon et al., 2001, 2004), Gross Motor Performance Measure (GMPMM) (Buckon et al., 2001, 2004), Berg Balance Scale (BBS) (Christovao et al., 2015; Zhang et al., 2009;), Bruininks-Oseretsky Test of Motor Proficiency (BOTMP) (Buckon et al., 2004), TUG (Christovao et al., 2015) and the six-minute walk test (Christovao et al., 2015).

Buckon et al. (2001) found that functional skills of the PEDI were significantly improved in all AFO configurations compared to barefoot ($p<0.004$) in those with hemiplegia, with no differences found among the three AFO configurations. When using the PEDI to assess those with diplegia, Buckon et al. (2004) reported that AFO use did not significantly improve mastery of functional motor skills or the level of independence.

Buckon et al. (2001, 2004) both investigated quality of movement using the GMPM in their studies. The researchers noted that there was significant improvement in motor performance in (quality) those with hemiplegia (Buckon et al., 2001) when wearing HAFO and PLS more so than SAFO, but no significant gain in gross motor function, whereas in those with diplegia (Buckon et al., 2004) found significant gains in gross motor function, but not performance. Zhang et al. (2009) found that after three months of treatment (rehabilitation and HAFOs intervention) BBS scores were significantly better than those in SAFO group ($p<0.01$). Christovao et al. (2015) did not find any difference in BBS when tested in PCI or control group placebo insoles. Buckon et al. (2004) found significant improvements in the BOTMP in all AFO configurations compared with barefoot in the upper limb coordination ($p<0.003$) and upper limb speed and dexterity ($p<0.016$) subtests. Christovao et al. (2015) noted that when the intragroup analysis was performed, significant differences were only found in the experimental group on the TUG when shoes with insoles were compared to shoes without insoles ($p=0.021$). There were also no differences noted in the six-minute walk test when comparing experimental group with barefoot or control group (Christovao et al., 2015).

3.4.6. Gait Parameters

Three studies (Buckon et al., 2001, 2004; Smith et al., 2009) used kinetic and kinematic gait outcome measures. Buckon et al. (2001, 2004) both measured gait, energy expenditure and functional motor skills between three different AFO designs, all of which were compared to a barefoot condition (except for energy expenditure which was compared with wearing shoes only). Smith et al. (2009) analysed gait metrics between barefoot and whilst wearing HAFO and DAFO.

Stride ($p<0.006$) (Buckon et al., 2004) ($p\leq0.05$) (Smith et al., 2009) and step length ($p<0.005$) (Buckon et al., 2004) were significantly increased ($p=0.0001$) (Buckon et al., 2001), and cadence was significantly decreased, ($p<0.002$) (Buckon et al., 2001) ($p<0.005$) (Buckon et al., 2004) in all three AFO configurations compared to barefoot. Smith et al. (2009) found a significant decrease ($p\leq0.05$) towards normal cadence were seen when the HAFO and DAFO conditions were compared with barefoot conditions. They also noted kinematic changes were most prominent at the ankle during brace wear, with minimal changes found at the hip and knee. One paper (Buckon et al., 2004) reports self-selected walking velocity was unchanged in any of the AFO configurations compared to barefoot, however another paper (Buckon et al., 2001) showed significantly increased velocity in the three AFO configurations compared to the shoes only condition ($p=0.0001$).

There were only two papers reporting on energy expenditure and oxygen consumption. One paper (Buckon et al., 2001) found during self-selected walking there were no significant differences among the four assessment conditions for oxygen consumption and energy cost, however another paper (Buckon et al., 2004) found energy cost was significantly decreased in self-selected and fast walking in all AFO configurations compared with shoes only ($p<0.003$). They also noted the HAFO maximized ankle power generation and improved the energy expenditure, whereas the PLS was the most effective in normalizing gait parameters.

3.4.7. Other Outcome Measures

Four studies (Buckon et al., 2001, 2004; Smith et al, 2009; Zhang et al., 2009) looked at ankle range of motion (ROM) and one assessed the Modified Ashworth Scale (MAS) (Zhang et al., 2009). One paper (Christovao et al., 2015) looked at static balance using force plate (Kistler, model 9286BA) and one study (Smith et al., 2009) looked at PODCI.

Buckon et al. (2004) did not find any significant change in lower extremity ROM but Buckon et al. (2001) found that ankle dorsiflexion knee extension was significantly increased in the HAFO and the PLS compared to barefoot ($p=0.003$); however, no significant change was found in the SAFO. Zhang et al. (2009) found that after three months of treatment (rehabilitation and HAFOs intervention) the MAS and ROM were significantly better than those in SAFO group ($p<0.01$). Christovao et al. (2015) found significant differences were found only in the experimental group on anteroposterior sway with eyes closed (shoes without insoles vs. barefoot, $p=0.050$) when intragroup analysis was performed. Smith et al. (2009) found there was no significant difference in the PODCI among barefoot and all AFO conditions ($p\leq0.05$).

Table 8: Details and summary of selected studies

Author	Design	Participants	Orthotic Intervention	Orthotic prescript	Timing of Assessment	Outcome measures	CAS (%)	Downs and Black, Grade
Cristovao et al. (2015)	Randomised, controlled, double-blind clinical trial	20 children with spastic diplegic CP, GMFCS I or II Experimental Group; m=2, f=8, age range 4-7.9 Control group; m=3, f=7, age range 5-9	Own AFOs, insoles (placebo or postural)	6 hours / day	Assessed at baseline (no orthoses), at 3-month period, one month after ceasing wear	BBS, Timed Up-and-Go Test, Six-Minute Walk test, GMFM-88	85	23 Good
Bjornson et al. (2006)	Randomized crossover design	23 children with spastic CP, (m = 52.2%), mean age 4.3years GMFCS I (6), II (3), III (14), current use of bilateral DAFO with free plantarflexion	Bilateral DAFO	30 min adaptation period	GMFM-88 assessed with and without DAFO, one 3 – hour visit	GMFM -88, correlations between demographics to change in gross motor skills	85	23 Good
Buckon et al. (2004)	Randomized controlled trial	16 independent ambulatory children, diplegic CP, (m=10, f=6) mean age = 8y 4mo, range 4 y4m to 11y6mo GMFCSI or II	Custom made SAFO, HAFO, PLS	6-12 hours wearing/day	4 visits over 12-month period, assessed at end of each wearing period. No usual physio over this time	Ankle passive range of motion, Energy Expenditure, gait analysis (kinematic / kinetics), Functional skills (BOT, GMFM-88, GMPM, PEDI)	67	18 Good
Smith et al. (2009)	Quasi-experimental with pre/post design	15 children with spastic diplegic CP (mean age 7.5±2.9years) GMFCS I, 20 children (normal development) as control group (mean age) 10.6±2.8years	DAFO, HAFO	1.Barefoot (baseline 1) 2. First AFO for 4 weeks 3. Barefoot after no AFO for 2 weeks (baseline 2)	Whilst barefoot (baseline 1), after wearing first AFO for 4-weeks, whilst barefoot after wearing no orthosis for 2 weeks (baseline	GMFM, PODCI, kinematic and kinetic gait analysis performed barefoot and with orthoses	67	18 Good

				4. after wearing alternative	2), after wearing the alternative AFO for 4weeks.			
Buckon et al. (2001)	Randomised controlled trial	30 children (21 male, mean age 9 years 4months; age range 5 years 3 months -15 years 3 months), able to walk without assistive devices	HAFO, PLS, SAFO	3-month baseline of no AFO wearing, 6-12 hours of AFO use (removed at night) over 3-month period, repeated for each orthoses (i.e., 1-year total)	Initial assessment after 3 months baseline, then after each 3-month period.	Ankle ROM, 3D gait analysis (kinetic and kinematic), energy consumption, GMFM, GMPM, PEDI	74	20 Good
Dalvand et al. (2013)	Quasi-experimental with pre/post design	30 children with spastic diplegic CP (m = 13, f = 17), age range 4-8years. GMFCS Levels I (12), II (13), III (5)	Custom made SAFO / HAFO	Wore AFO regularly for 3 months, 6 hours / day plus attend 36 OT training sessions Control group who only had OT training for 3 months	GMFM-88 assessed pre- and post-orthotic prescription and use as described 3 months (in between assessments)	GMFM-88 either side of orthotic prescription	85	23 Good

Zhang et al. (2009)	Randomized crossover design	38 children with spastic diplegia (21-71 months), able to walk	HAFO, SAFO	3 months, wearing orthoses 6-12hr/day, rehabilitation training 1hr/day, 5x/week for 3 months.	Initial barefoot balance baseline assessed. Beginning and end of 1st month and 3rd month all outcome measures assessed.	Modified Ashworth Scale, ROM, BBS and GMFM-88	70	19 Good

AFO=Ankle Foot Orthoses: BBS=Berg Balance Scale: BOT=Bruininks-Osteresky Test: CP=Cerebral Palsy: Dynamic-AFO=Dynamic Ankle Foot Orthotics: GMFCS=Gross Motor Functional Classification System: GMFM-88=Gross Motor Function Measure: GMPM: Gross Motor Performance Measure: Hinged-AFO=Hinged Ankle Foot Orthotic: PEDI=Pediatric Evaluation of Disability Inventory: PLS=Posterior Leaf Spring: PODCI=Pediatric Outcomes Data Collection Instrument: Range of Motion = ROM: Solid-AFO=Solid Ankle Foot Orthotic

Table 9: The results of the critical appraisal of included studies

Authors	Study design (EBMLE) (Howick, et al., 2011)	Reporting (/10)	External validity (/3)	Internal validity – bias (/7)	Internal validity – confounds (/6)	Power (/1)	Total (/27)	CAS (%)
Cristovao et al. (2015)	Randomized Controlled Trial (Level 1b)	9	3	7	4	0	23	85
Bjornson et al. (2006)	Prospective, randomized crossover design (Level 2b)	9	3	7	4	0	23	85
Buckon et al. (2004)	Randomized Controlled Trial (Level 1b)	8	3	5	2	0	18	67
Smith et al. (2009)	Quasi-experimental Study (Level 4)	6	3	5	4	0	18	67
Buckon et al. (2001)	Randomized Controlled Trial (Level 1b)	8	3	5	4	0	20	74
Dalvand et al. (2013)	Quasi-experimental Study (Level 4)	9	3	7	4	0	23	85
Zhang et al. (2009)	Prospective, randomized crossover design (Level 2b)	8	3	5	3	0	19	70
	Mean (SD) Range	8.0±1.0 6-9	3 -	5.7±1.0 5-7	3.5±0.8 2-4	0 -	20.1±2.3 18-23	74.6±8.5 67-85

CAS = Critical Appraisal Score: EBMLE = Evidence-Based Medicine Levels of Evidence

Table 10: The statistical analysis, results, strengths and limitations of included studies

Author	Statistical analysis	Results	Strengths	Limitations
Cristovao et al. (2015)	- 2 -way ANOVA (Bonferroni post hoc test)	- Timed Up-and-Go significantly (*p*=0.021) better results in experimental group vs control - Experimental group demonstrated significantly (p=0.050) reduced body sway (improved static balance) compared to control group - No carryover benefit after cessation of postural control insole use	- Highest overall Downs and Black score - Randomised Control Trial - Many clinically relevant OMs	- Slightly reduced internal validity and confounding
Bjornson et al. (2006)	-Correlations; Wilcoxon signed rank test, Secondary analyses used, Spearman rank correlation test, Kruskal-Wallis test, Mann-Whitney test -p value - mean ± SD	- GMFM-88 % scores significantly higher (*p*>0.001), when wearing DAFO vs none - GMFM-88 standing dimension change was negatively correlated with age (r= -0.21) - Average leg length correlated with total GMFM and W/R/J - significant (*p*=0.005) change in standing, W/R/J and total GMFM-88 with independent walkers' vs aided.	- Highest overall Downs and Black score - Randomised crossover trial - Strong reporting - Strong internal validity (bias) - Strong external validity - No dropout or adverse events - Appropriate assessments and analyses of results	- Poor internal validity (confounding) -Only comparing DAFOs to barefoot
Buckon et al. (2004)	- Multiple one-way repeated ANOVA - Bonferroni corrections - Paired t-test - Mean ± SD	- AFO use enhanced functional abilities of most children with spastic diplegia - Regardless of configuration, AFO did not significantly alter pelvic and hip kinematics and/or kinetics - HAFO significantly (*p*>0.007) increased peak knee extensor moment in early stance - All configurations significantly altered ankle kinematics during the stance / swing phases of gait: DF at initial contact (*p*=0.0001), peak DF in stance (*p*<0.009), timing of peak DF in stance (*p*<0.003), peak DF in swing (*p*<0.0002), dynamic ankle range (*p*<0.0001), increased step length (*p*>0.005), stride length (*p*>0.006), Upper Limb Coordination (*p*>0.003), speed and	- Very detailed results and reporting - Multiple OM demonstrating varying changes	- External validity and power -Internal validity - Confounding - Lowest score overall in Downs and Black

		dexterity (p>0.016), dynamic dimension of GMFM-88 (p>0.002), decreased cadence (p>0.005) and energy cost (p>0.003).		
Smith et al. (2009)	- Multiple one-way repeated ANOVA - Bonferroni corrections - Multiple regression (Fourier series) - Wilcoxon signed rank sums - Mixed-effect linear model with random effect - p value	- Brace wear demonstrated significant improvements in gait metrics - No significant differences seen between two braces used - Barefoot and braced conditions were found to have similar scoring with PODCI and GMFM sections D and E	- Randomised bracing sequence - Strong external validity	- Lowest level of reporting per Downs and Black
Dalvand et al. (2013)	- Wilcoxon signed rank test, Kruskal-Wallis test, Mann-Whitney U test	- HAFO significantly improves gross motor function over SAFO (p>0.05 and control groups (p>0.01) - gross motor function improved in all groups	- Quasi-experimental design - 30 participants - Random assignment into groups -Strong overall Downs and Black results	- Small sample size - Low tech clinical tools - Poor internal validity- confounding
Buckon et al. (2001)	- mean ± SD - one-way repeated ANOVA - Bonferroni (with p value significance) - linear contrasts	PROM; - DF with knee extended significantly increased (HAFO, PLS) compared to barefoot (SAFO insignificant), nil changes with knee flexed GAIT ANALYSIS; - Significantly increased in all AFO compared to barefoot (p=0.0001); initial contact and stance, stride and step length. - Significantly greater; initial contact in HAFO and SAFO (p=0.002, p=0.015) compared to PLS, stance in HAFO compared to PLS and SAFO (p=0.004, p=0.0001). -Stance peak ankle power generation significantly decreased in all AFO configurations compared to barefoot (p=0.003 HAFO,	- Long timeline of study - Used baseline - 30 participants - very detailed reporting and results -multiple OMs reporting change	- Low internal validity - bias

		p=0.001 PLS, *p*=0.0001 SAFO). Significantly less in SAFO compared to PLS and HAFO (*p*=0.001, *p*=0.0001). - cadence significantly decreased (p<0.002) in all three AFO configurations compared to barefoot. ENERGY CONSUMPTION; - nil changes when self-directed walking or fast walking. HAFO significantly decreased energy cost (shoes on; *p*=0.002). GROSS MOTOR / FUNCTIONAL SKILLS All AFO configurations significantly improved WRJ dimension of the GMPM (*p*<0.005). Alignment and Stability were not improved by AFO use, Coordination and Weight Shift significantly improved by the HAFO (*p*<0.004, *p*<0.004, PLS (*p*<0.005, *p*<0.0001) and SAFO (*p*<0.005, *p*<0.0002) compared to barefoot. -PEDI; Functional skills were significantly improved in all AFO configurations compared to barefoot (*p*<0.004) with no differences found among the three AFO configurations		
Zhang et al. (2009)	-% -p value - mean ± SD	- Noted OMs and function changes noted after AFO use, with HAFO more significant (*p*<0.01) than SAFO - HAFO can correct foot drop, increase muscle strength of DF when walking	- Random assignment of orthoses - Rehabilitation as additional intervention - Many clinically relevant OMs - Allowed wear in time for orthoses -38 participants	- did not report precise *p* value

AFO=Ankle Foot Orthotics: ANOVA=Analysis of variance: Dynamic-AFO=Dynamic Ankle Foot Orthotics: DF=Dorsiflexion: GMFCS=Gross Motor Functional Classification System: GMFM-88=Gross Motor Function Measure: GMPM: Gross Motor Performance Measure: Hinged-AFO= Hinged Ankle Foot Orthotic: OM=outcome measure: PF=plantarflexion: Solid-AFO=Solid Ankle Foot Orthotic: SD=standard deviation: W/R/J=walking/running/jumping

3.5. Discussion

The aim of this review was to identify and critically analyse literature investigating the use of lower extremity orthoses in children with CP and to synthesize the findings. The hypothesis that lower limb orthoses would improve alignment, cadence and balance was confirmed. The search strategy retrieved seven fair to good quality studies, all noting improvement in gait and GMS when wearing AFOs.

Dalvand et al. (2013) found an improvement in gross motor function amongst all groups (p<0.01), however HAFO demonstrated a greater improvement than SAFO and control groups. This improvement was thought to be secondary to the ability of the HAFO to normalize ankle motion by limiting ankle plantarflexion and allowing for free dorsiflexion in the stance phase. Buckon et al. (2001) reported similar findings being the majority of children with spastic hemiplegia demonstrated greatest benefit in enhanced functional motor skills with either HAFO or PLS. However, Buckon et al. (2004), noted a subtle but detrimental effect on function with HAFO use, recommending that some children with spastic diplegia (GMFCS II) should have constrained ankle motion by using PLS or SAFO.

Buckon et al. (2001) did not recommend SAFO for the management in children with hemiplegia, instead recommended a more dynamic orthosis such as the PLS or HAFO. Smith et al. (2009) report no significant differences between barefoot and orthoses for sections “D” and “E” of the GMFM-88 but report that DAFOs improved gait parameters, although this may be due to all participants being GMFCS I.

Bjornson et al. (2006) compared DAFO to shoes alone and reported that children who were independent walkers appeared to benefit greater from the DAFO with free plantarflexion in comparison to those children who require assistive devices (types not reported) to mobilise. The authors found the GMFM percentage scores for all dimensions were significantly higher (p<0.001) with the children wearing DAFOs during same day evaluations. Conversely, Buckon et al. (2004) that most (but not all) children should be considered to have their ankles constrained by using a PLS or SAFO.

Buckon et al. (2004), noted that the AFOs did not significantly improve standing skills of the GMFM-88 (p≤0.025), but reported an improvement of the walking, running and jumping elements when wearing AFOs (p<0.002) compared to barefoot. Zhang et al. (2009) found similar outcomes reporting that HAFO is superior to SAFO in improving standing, running and balance function. Buckon et al. (2001) noted that despite the AFO enhancing stability throughout static and dynamic gross motor and functional skills, they did not allow the child to achieve a skill they were previously unable to master. They also note that some aspects of section "D" of the GMFM-88 require transitions into standing, which may be difficult for those children in AFOs and may account for the decrease in function when using AFOs. Christovao et al. (2015) found that postural insoles led to an improvement in dynamic and static balance among children with CP, as demonstrated by the improved TUG and reduction in body sway in the anteroposterior and mediolateral directions.

Zhang et al. (2009) found that wearing SAFO contributed to 33.3% of the children developing plantar muscle atrophy, heel dysplasia and weakened flexor or toe active movement when compared to those wearing the HAFO. Zhang et al. (2009) also recommended different AFO types be used depending on the specific needs and circumstances of the child. Zhang et al. (2009) noted as SAFOs have advantage in stabilizing the ankle joint which benefits hip and knee extension, they should therefore be used in cases of increased plantarflexion, knee flexion and reduced muscle strength. Zhang et al. (2009) noted, however, that HAFO have advantage when there is more movement into dorsiflexion, whereby the second rocker motion is facilitated, and the knee controlled throughout gait cycle. The authors supported the use of both orthoses as the functional benefits include stabilisation and support, prevention and correction of deformity, reduction of axial load bearing, inhibition of muscle tension during standing and walking, and improvement of walking ability (Zhang et al., 2009).

The volume of evidence trending throughout all seven selected studies suggests that wearing an orthotic device does improve the gait and GMS of children with CP. However, the inconsistency in recommendations of design, use, and prescription leads to an unstable foundation on which therapists and medical professionals must base their clinical and evidence-based reasoning. There is also no recommendation of how to adapt prescription or LLO structure to enhance the user's ability in all areas of GMS, and to address the limitations found.

While several types of orthoses were discussed across the included studies, there were no apparent studies completed using the SMotOs in children with CP. The most similar description of orthoses was the study by Christovao et al. (2015). The PCIs described portray a similar orthotic to SMotO as they included the description of the raised areas on the sole of the orthotic and worked under a similar theory; affecting sensorial afference and stimulate a postural reaction but did not appear to describe specific foot postural muscles as those noted in papers utilizing SMotO (Mabuchi et al., 2012; Subke et al., 2009). Their findings suggested that PCIs affect sensorial afference and stimulate a postural reaction, favoring better biomechanical alignment of the body and permitting more efficient function. Mabuchi et al. (2012), investigated the biomechanical effect of the SMotO when utilised in children with an in-toeing gait. They concluded that SMotO improved the in-toeing gait pattern by externally rotating the proximal femur during end stance. Along with poor biomechanical alignment, they also demonstrated the effective use of SMotO on a population with high tone gastrocnemius, which could provide some basis for evidence for use in CP. Subke et al. (2009) investigated the optimization of the use of the SMotO and provided the first steps towards a basis for foot-insole-interaction via numerical analysis, thus providing a standard for prescription. These three papers indicate a promising value in further investigation of SMotO and PCIs in children with CP. As such, there appears to be a lack of identified research investigating the use of SMotO in this population and further research employing this specific type of orthosis is needed.

There was, however, one key limitation noted in the literature, although thorough search strategies were implemented, there were no papers were found using SMotOs in this population. Despite sound reasoning on the physiological mechanism of how SMotOs work, and its use in clinical practice, there is little English based peer-reviewed research available. Due to its simplistic form, ease of use, clinically relevant physiological response, this orthosis may be a viable contender for use in CP population where appropriate.

A strength of this review entailed the use of two researchers (CM, VS) to independently perform the critique of the papers and high subsequent level of agreement. This approach was aimed to reduce the possibility of bias and increase the consistency in reliable evaluation. This review assessed three RCTs. Due to the processes involved during the conduct of an RCT, the risk of confounding factors influencing the results was minimised, meaning the RCT is considered to provide the most reliable level of evidence (Akobeng, 2005) as evident by the higher EBMLE score (1b) and CAS (85%) given in this review. Another strength was the use of a paper translated from Chinese (Zhang et al., 2009) demonstrating a wider range of research location. Furthermore, using the modified Downs and Black checklist ensured the volume of evidence reviewed in this review was associated with the quality of the research used to establish this evidence. According to the Kennelly (2011) grading scores, the seven included papers were all of good quality, therefore demonstrating a strong search strategy to this review. Finally, the last strength of this review was the numerous databases included in the search strategy.

3.6. Conclusion

Overall, the literature supported the use of AFOs as a device to improve gait and GMS in children with CP. However, there was inconsistency in the agreement of design and use. Given the findings of this review on the variations in orthotic device prescription, due in part to the variability in requirements of children with CP, there is a potential benefit to the clearer defining of AFO prescription within research studies.

In addition, the lack of published research investigating the use of sensomotoric orthoses, specifically with regard to gait and GMS in children with CP, provided further motivation to explore the effect of sensomotoric orthoses on gait and GMS in children with CP in the following studies.

4.0. Feasibility & Pilot Study: Investigating Feasibility of Using Multiple Outcome Measures in Children with CP When Wearing Sensomotoric Orthoses & Ankle-Foot Orthoses

Prelude

The following chapter continues the theme of outcome measures determining how lower limb orthoses improve gait and gross motor skills in children with cerebral palsy. As ankle-foot orthoses have been found to be a successful intervention (Chapter 3) in addressing gait and gross motor skills in children with cerebral palsy, this pilot study will continue the theme of orthoses. This chapter will detail the study which investigated the feasibility of using multiple outcome measures to assess the effect of two orthoses (sensomotoric orthoses and ankle-foot orthoses) on gait and gross motor skills in children with cerebral palsy. This pilot study used outcome measures described in the systematic narrative review (Chapter 3) that were found to be clinically valid and reliable for use in a population of children with cerebral palsy. Chapters 4, 5 and 6 are subsections using the same participants.
See Appendices D though F for full evidence and protocols for outcome measures used.

4.1. Introduction

Improving gait and gross motor skills tend to be a frequent and primary goal of therapy in children with cerebral palsy (CP) (Alotaibi, Long, Kennedy, & Bavishi, 2014; Thomai & Gita, 2017). Through evaluation, therapists are able to address the areas of deficit. Gait analysis appears to be one of the important aspects of a therapist's evaluation (Gupta & Raja, 2012), as well as accurate measurement of changes in gross motor skill acquisition to determine the effectiveness of intervention programs (Alotaibi et al., 2014). A literature review by Craig (1999) echoes the general difficulties encountered by physiotherapists in the application of outcome measures (OM) within clinical practice, where time is limited and ease of application a necessity often underemphasized in the literature. OMs can be used to determine the changes brought about by an intervention.

Similar to the postural control insole (Christovao et al., 2015), sensomotoric orthotics (SMotO) provide a different approach than the usual AFO to the management of gait and spasticity in children with CP. Wegener, Wegener, Smith, Schott, & Burns (2016) described one adaptation theory as 'elements' on the foot orthoses that increase or decrease local pressures detected by cutaneous receptors, muscle spindles or Golgi apparatus in order to reduce the activity of over-active muscles, which in turn facilitates an increase in the activity of weaker muscles (Ludwig, Quadflieg, & Koch, 2013; Ohlendorf, 2013; Subke, Kolling, Griesemann, Kleinau, & Staudt, 2009). Given the physiology of ascending muscle chains, the reaction will influence the complete chain of movement (Ohlendorf, 2013). Clinically, these orthoses have been used with children with CP and observed to improve gait and gross motor skills (GMS).

To demonstrate the impact of an intervention, such as an ankle-foot orthosis (AFO), or SMotO on gait and gross motor skills in children with CP, appropriate OMs can be applied. As a role of a paediatric physiotherapist is to provide successful intervention, the effectiveness of said intervention must be assessed. Through using valid and reliable OMs for children with CP, physiotherapists can accurately monitor the effects of intervention and modify therapy as such.

As yet, there are no studies using Gross Motor Function Measure (GMFM), Berg Balance Scale (BBS), Timed Up-and-Go (TUG), Edinburgh Visual Gait Score (EVGS) and Cerebral Palsy Quality of Life (CPQoL) to report the effect of SMotOs and

AFOs on gait and gross motor skills (GMS) in children with CP. Due to limitations of available research, this pilot study was conducted to determine the feasibility and assess the ability to collect a variety of OMs validated for use in children with varying levels of CP and severity. It was hypothesised that using three outcome measures (twice) with two separate lower limb orthoses would be feasible to implement in a timely manner. Therefore, the aim of this pilot study was to determine the feasibility of using multiple OMs that assess changes in gait and gross motor skills in children with varying levels and presentations of CP when in AFOs and SMotOs. The results will inform further studies investigating the effect of SMotOs vs AFOs on gait and GMS in this same population.

4.2. Methods

This pilot feasibility study was a mixed method study design, with both objective and subjective OMs collected.

4.2.1. Participants

4.2.1.1. *Inclusion Criteria*

The participant inclusion criteria were a) a diagnosis of CP, b) no surgery in past six weeks, and c) currently using SMotO (completed the wearing in process of at least two weeks).

4.2.1.2. *Recruitment*

Participants were children with CP recruited via convenience sampling, through two private therapy practices (Therapies for Kids and NAPA Centre, Sydney, Australia).

4.2.2. Intervention (orthoses)

All participants brought their own SMotOs and AFOs to the data collection sessions. The AFOs were all custom made through either public hospital or private orthotist and were made from polypropylene with Velcro straps holding the foot in place. The SMotOs were custom made for each child from ethylene vinyl acetate (EVA) and had been assessed and prescribed by a podiatrist or pedorthist who were both expert in this design type of orthosis.

4.2.3. Setting

Testing was undertaken in two separate clinic locations, as well as home settings when families were unable to travel. These settings were familiar to the child, with parents and/or siblings present throughout the testing (where applicable).

4.2.4. Outcome Measures

Only clinically relevant and repeatable OMs were selected. The OMs with 50% participation rate or higher will be considered as viable to include in the future studies. The OMs performed by children in both the SMotO and AFO were the TUG, BBS, GMFM-88 and EVGS. The CPQoL was taken home by families to complete and return. Please refer to the Appendix D (i to vi) for full details of each OM undertaken.

4.2.4.1. Minimal Clinically Important Difference

The minimal clinically important difference (MCID) for the BBS was noted by Gervasoni, Jonsdottir, Montesano, and Cattaneo (2017) as three points of difference in the final score. Robinson et al. (2017) reported the MCID for the EVGS was 2.4. According to the GMFM-88 user manual system, a percent increase of 5-7% of score is classed as a medium positive change (CanChild, 2020). The Minimal Detectable Change (MDC) and MCID for the TUG in children with CP, being: GMFCS Level I: 1.40 seconds, GMFCS Level II: 2.87 seconds, and GMFCS Level III: 8.74 seconds (Carey, Martin, Heathcock, & Comb-Miller, 2015). The MCID and MDC was used as a reference point for the results across this study.

4.2.5. Process

Ethical approval was obtained through the Bond University Research and Ethics Committee (Approval RO-1835). Consent was gained from Clinic Directors in both private practice settings. Participants and their caregivers were given an explanatory statement and consent form, both of which were read and completed before data collection took place. The primary researcher followed the individual protocols of each outcome measure, which were assessed in both the AFO and SMotO. The child had a small break in between orthotics. The parent / caregiver was given the CPQoL (Appendix D, Section v) to complete and return to the researcher.

4.3. Results

There was a total of six participants with CP (male: n=5 male; female: n=1) who were aged between 3 - 13 years old. All participants were diagnosed with CP, with a range of Gross Motor Function Classification System (GMFCS) I, II & IV. The types of CP were inclusive of; spastic quadriplegic CP (n=3), dystonic spastic quadriplegic CP (n=1) and spastic diplegic CP (n=2).

Of the six children, one (16.7%) completed the CPQoL, three (50%) completed the BBS, one (16.7%) completed the TUG, four (66.7%) completed the GMFM-88, and six (100%) took part in the gait analysis. Factors affecting participation in each area of assessment included; unable to follow verbal direction (for GMFM-88, BBS and TUG), unable to stand without support (BBS), did not bring walker (TUG), too young, incomplete or failure to return questionnaires (CPQoL). Refer to Table 11 for full details.

Table 11: Participants and their outcome measures

Outcome Measure / Participant	BBS	TUG	GMFM-88	EVGS	CPQoL	TOTAL /5
1	Y	x	Y	Y	x	**3**
2	Y	x	Y	Y	x	**3**
3	Y	Y	Y	Y	x	**4**
4	x	x	x	Y	Y	**2**
5	x	x	Y	Y	x	**2**
6	x	x	x	Y	x	**1**
TOTAL /6	**3**	**1**	**4**	**6**	**1**	

BBS = Berg Balance Scale; TUG = Timed Up and Go; GMFM-88 = Gross Motor Function Measure; EVGS = The Edinburgh Visual Gait Score; CPQoL = Cerebral Palsy Quality of Life Questionnaire

Only one participant out of six completed the TUG. This was due to reasons such as reduced comprehension (n=1), poor behaviour (n=1) or physical inability to complete the task (n=3). The BBS had 50% participation rate due to reasons such as requiring equipment to maintain standing or balance (n=1), poor comprehension of task required (n=1), poor behaviour and attention (n=1).

There were three outcome measures that had the highest compliance and completion rate – BBS, GMFM-88 and EVGS. This may have been due to factors such as ease of use for clinician, reduced instruction comprehension required from participant, outcome measures appropriate for all levels of physical ability.

4.4. Discussion

The hypothesis that using three outcome measures (twice) with two separate lower limb orthoses would be feasible to implement in a timely manner was confirmed. Overall, there were six participants who were able to attend data collection sessions and participate in two or more outcome measures. All levels of GMFCS were accepted into the study as the researchers did not want to discriminate against higher or lower functioning children. It was also deemed that participation and feedback from parents of these children with a higher level GMFCS was important as it appeared there was not a lot of research on effect of orthoses at higher severity of disability. Throughout the outcome measure data collection, it was noted that the tasks that required less intellectual comprehension, with simpler instructions or physical tasks had the higher participation rate. It was also noted by the researcher that when the child was tiring, the participation level reduced, and it was harder to keep on task.

Five participants did not complete the TUG. Due to only one participant completing this outcome measure, the TUG was determined by researchers not to be a consistently viable outcome measure for this specific population group. The BBS had three unable to complete the OM and was also removed from consideration for including in further studies. The CPQoL was deemed to be not feasible as an assessment tool in this setting as many families were unable to complete all the answers, did not send the questionnaire back, or made errors in their marking therefore were deemed incomplete.

Overall, the feasibility of assessing five total outcome measures was determined to be ineffective due to; time taken to complete several outcome measures, inability of the parent to commit to timeline, fatigue of the child, non-compliance to instruction, inability to comprehend instructions, inability to perform physical tasks, inability to complete outcome measures and behavioural issues.

Conversely, there were three successful OMs: the GMFM-88 (66.6%), the EVGS (100%) and the BBS (50%). The researcher reported that the GMFM-88 was relatively easy to use to assess the participants as some of the tasks were able to be completed without verbal, receptive or expressive language, rather, the participant was cued through motivation to reach for a toy or was placed in a position to assess movement out of the position. Many of the participants appeared comfortable with the application and sequence of this outcome measure as it had been seen in assessment sessions conducted through other therapists in the past. There were two participants who were unable to complete the GMFM-88 due to ability to follow direction due to behaviour and ability to follow direction.

Time wise, the EVGS had many benefits for the researcher as the participant was filmed and gait data extracted and analysed at a later date. It also did not require a high level of comprehension from the participant. The BBS had the lowest participation rate at 50% but was still included as it achieved the minimum participation rate.

There was a small but clear trend for increased quality of movement with the BBS and TUG. However, the limitation of low participant numbers affected the significance of participation.

4.4.1. Limitations

There were a small number of recruited participants (n=6) which may affect the significance of the results. Many of the participants failed to complete the full selection of outcome measures which may suggest that the participant's families were not adequately aware or committed to their responsibility as a participant in the study. This may also be due to the complexities of having a child with a disability, and their priorities may not have aligned with the study at that time. Examples include only one participant completing the CPQoL, and one participant completing the TUG test. Another limitation was this study did not assess how effective the OMs were at measuring alterations in gait, therefore future studies could investigate the link between practicality of use and the ability of the OM to measure an actual change.

4.5. Conclusion

There were three out of six outcome measures that demonstrated viable results through participant response and analyses that will be used in further, in depth studies relating to the effect of orthoses on children with CP. The BBS, GMFM-88 and EVGS will be used in following studies to demonstrate the changes in gait and gross motor skills in children with CP when wearing two different types of orthoses. A parent focused questionnaire will be created to further drive a subjective focus on the use of two types of orthoses and how this affects quality of life for the family and child.

5.0. Effect of Sensomotoric Orthoses & Ankle-Foot Orthoses on Gross Motor Skills in Children with Cerebral Palsy

Prelude

The following study reported in this chapter uses the selected outcome measures from the previous chapter (Study 4) to assess the effect of orthoses on gross motor skills in children with cerebral palsy.

5.1. Abstract

Background: Cerebral Palsy (CP) is a neurodevelopmental condition well recognised to begin at birth or early childhood. Ambulatory aids and lower limb orthoses are a frequently prescribed intervention for children with CP (Russell & Gorter, 2005). Ankle-foot orthotics (AFOs) are the typical prescription of lower extremity orthoses for the management of lower limb deformities that often occur with CP. Unlike AFOs, sensomotoric orthotics (SMotOs) provide a different approach to the management of gait in children with CP. Clinically, the SMotOs have demonstrated improved foot alignment, balance, control with walking and functional skills. **Intervention:** The Gross Motor Function Measure (GMFM-88) and Berg Balance Scale (BBS) are two examples of instruments that can be used to evaluate change in gross motor function in children with CP. The aim of this study was to compare the effects of SMotOs and AFOs on gross motor skills, assessed by GMFM-88 and the BBS in children with CP. **Methods:** Participants were children with CP, using both SMotO and AFO, recruited from two private practice clinics in Sydney and performed the GMFM-88 and BBS in both orthoses. **Results:** Nine participants took part in this study (aged between 3 – 13 years with average 5.4 ± 3.2 years) who had spastic diplegia (n=4) and spastic quadriplegia (n=5). Participants wore hinged-AFOs (n=4) and solid-AFOs (n=5). On average, participants scored significantly (p=0.002) higher on the GMFM-88 when wearing SMotOs compared to AFOs. The total mean BBS score for SMotOs was 22.00 (±13.00) points and for AFOs was 21.88 (±15.37) points. There was no significant difference between BBS total scores when wearing SMotOs and AFOs (p=0.928). **Conclusion:** The results demonstrated an effective and significant benefit on gross motor skills when wearing SMotOs compared to wearing AFOs when assessed using the GMFM-88.

5.2. Introduction

Cerebral Palsy (CP) is a neurodevelopmental condition well recognised to begin at birth or early childhood and persisting through the lifespan and is the leading cause of permanent physical disability in children (Alotaibi, Long, Kennedy, & Bavishi, 2014; Ko & Kim, 2013; Paulson & Vargus-Adams, 2017; Rosenbaum, Paneth, Leviton, Goldstein, & Bax, 2007). It has an incidence of 2.0 to 2.5 per 1000 live births (Graham & Selber, 2003).

CP affects the development of movement, postures and causes activity limitation (Christovao et al., 2015; Ridgewell, Dobson, Bach, & Baker, 2010; Rosenbaum et al., 2007). It is characterised by abnormal motor patterns and postures, with a variety of presentations (Graham & Selber, 2003), such as spasticity, dystonia, muscle contractures, weakness and difficulty in co-ordination, ultimately affecting control of movements (Paulson & Vargus-Adams, 2017). Spasticity is a term that describes a hypersensitive stretch reflex caused by damage to the pyramidal tract, responsible for transferring motor control signals from the brain to the spinal cord (Ries, 2017). Spasticity is prohibitive to intentional motion as any agonist muscle motion is often counteracted by a “spastic catch” of the antagonist muscle as it is being stretched (Ries, 2017). The neurological impairments result in physical activity limitations and potentially affect a child’s opportunity to perform daily tasks as well as affecting their lifelong participation in activities.

Effective, contemporary intervention programs are based around promoting function throughout the course of life. Rehabilitative, therapeutic and medical interventions available are aimed to improve independence in daily activities through improving mobility, motor performance and overall fitness. A primary goal of therapeutic intervention in children with CP is to promote gross motor skill performance as an essential component of functional mobility (Alotaibi et al., 2014).

Ambulatory aids and lower limb orthoses are a frequently prescribed intervention for children with CP (Russell & Gorter, 2005). A consensus conference of the International Society for Prosthetics and Orthotics identified the aims of lower extremity orthotic management in children with CP as to 1) correct and/or prevent deformity, 2) provide a base of support, 3) facilitate training of motor skills, and 4) improve efficiency of walking (Morris & Condie, 2009). Lower limb orthoses assist therapeutic intervention by increasing balance and support to enhance standing and walking (Russell & Gorter, 2005). Ankle-foot orthotics (AFOs) are the typical prescription of lower extremity orthoses for the management of lower limb deformities that often occur with CP. The goal of each AFO prescribed for a child with CP is the collective improvement of these biomechanical variables to increase the ease of taking an individual step with the potential to enhance walking activity and functional skills (Bjornson et al., 2016). AFOs are designed to; affect body structure (Wingstrand, Hägglund, & Rodby-Bousquet, 2014), support normal joint alignment and mechanics, provide variable range of motion (ROM) when appropriate, facilitate function (Brodke et al., 1989; Knutson & Clark, 1991; White, Jenkins, Neace, Tylkowski, & Walker, 2002; Wingstrand et al., 2014), stabilize the ankle / foot complex (Buckon et al., 2004) and enable a continuous Achilles / gastrocnemius stretch (Boyd, Pliatsios, Starr, Wolfe, & Graham, 2000; Hainsworth, Harrison, Sheldon, & Roussounis, 2007; Morris, 2002).

Along with joint alignment, other improvements that may be seen through the use of AFOs are the improvement in walking efficiency (Morris & Condie, 2009; Rethlefsen, Kay, Dennis, Forstein, & Tolo, 1999), position the foot for function (Kane, Musselman, Manns, & Lanovaz, 2016) and improvement in gait function and pain prevention (Westberry et al., 2007). Common types of AFOs seen in the literature are solid-AFO (SAFO), hinged-AFO (HAFO), and dynamic-AFOs (Buckon et al., 2001, 2004; Dalvand et al., 2013).

Unlike AFOs, sensomotoric orthotics (SMotO) provide a different approach to the management of gait in children with CP. Wegner, Wegener, Smith, Schott and Burns (2016) describe one adaptation theory as 'elements' on the foot orthoses (e.g., forefoot valgus posting or lateral rearfoot padding) increase local pressures which are detected by cutaneous receptors, muscle spindles or Golgi apparatus. To expand this explanation, SMotOs are created to directly change the muscle length (Subke, Kolling, Griesemann, Kleinau, & Staudt, 2009) and 'activate and deactivate' muscles by increasing or decreasing individually placed point specific pressure on musculotendinous structures in the foot of the tibialis posterior, peroneus brevis and the lumbricals/quadratus plantae. This theory implies the information that is transmitted by the sensors for the control of muscle activity is changed (Ohlendorf, 2013). Depending on these individual pressure bumps' height and placement, the muscles can be activated or restricted (Ohlendorf, 2013; Subke et al., 2009). CP affects the different areas of the brain, therefore interrupting signals sent to the muscles. The SMotOs work on the idea that the signals are being sent from the muscles back up to the spinal cord through activation of the Golgi bodies, therefore signalling muscles to respond to stimulus (Wegener et al., 2016).

Clinically, the SMotOs have demonstrated improved foot alignment, balance, control with walking and functional skills. Clients have been prescribed the SMotOs as a supplementary lower limb orthosis when it is noted they are finding functional movement restricted in the AFO. SMotOs have also been prescribed for children who require more feedback from their feet, where wearing shoes alone has not been effective. To date, there are no known studies on the use of SMotOs in children with CP.

An evaluative measure is used to document change within subjects over time to determine the effectiveness of treatment or to monitor the natural development of a condition (Boyce et al., 1991). The Gross Motor Function Measure (GMFM-88) is an example of an instrument that can be used to evaluate change in gross motor function in children who have cerebral palsy (Boyce et al., 1991). CanChild, a research centre that focusses on developmental conditions in children and youth, describes the GMFM-88 as an 88-piece assessment tool designed and evaluated to measure changes in gross motor function over time or with intervention in children with CP (CanChild, 2020). The GMFM-88 provides descriptive information about motor function for very young children or children with complex motor disability, such as those functioning in Gross Motor Function Classification System (GMFCS) level V, as it has items that describe early motor skills (CanChild, 2020). A longitudinal study by Russell and Gorter (2005) found that the GMFM-88 was sensitive to functional changes as a results of using an aid and/or orthoses. The validation sample for the original GMFM included children five months to 16 years of age (CanChild, 2020). Ko and Kim (2013) report the relative reliability of the GMFM-88 in children with CP as excellent (intraclass correlations [ICC] = .952-1.000).

Balance strategies of children with CP are different than the strategies of children without neurological impairments as they demonstrate increased co-contractions and poor distal to proximal pattern of muscle activation (Kembhavi, Darrah, Magill-Evans, & Loomis, 2002). As balance is an integral part of gross motor skills, the ability to measure balance function can be an important to determine intervention. One such tool used to assess balance is the Berg Balance Scale (BBS). The BBS is a 14-section test that covers balance challenges that the assessed may face in day-to-day activities (e.g., static standing without holding on, getting up and down from a chair, picking an object up off the floor). Kembhavi et al. (2002) determined the validity of the BBS in 36 children with cerebral palsy, age of eight to 12 years old versus children without health problems as control group. The BBS and GMFM-88 were applied to all children and researchers found the BBS can be considered as a valid clinical measure of balance for children with CP.

There are multiple studies assessing and comparing the motor changes in gait and other gross motor skills using different types of AFOs in children with CP (Buckon, et al., 2001, 2004; Dalvand, et al., 2013; Zhang, Wang, Yang, Zhao, 2009). Buckon et al. (2001) noted that despite the AFO enhancing stability throughout static (e.g., standing) and dynamic (e.g., walking) gross motor and functional skills, the AFOs did not allow the hemiplegic child to achieve a skill they were previously unable to master. In another study, Buckon et al. (2004), noted that the AFOs did not significantly improve standing skills of the GMFM-88 ($p \leq 0.025$), but reported an improvement of the walking, running, and jumping elements when children with diplegic CP were wearing AFOs ($p < 0.002$) compared to being barefoot. Dalvand et al. (2013) also noted improvements in gait with both SAFO and HAFO, although HAFO demonstrated better improvements in gross motor function than SAFO. From this research, the volume of evidence suggests AFOs are effective at improving gross motor and functional skills in children with CP.

However, there is a lack of evidence examining the ‘sensorimotor response’ paradigm, as there are no randomized trials, minimal peer-reviewed papers in English and only a few small cross-sectional paediatric papers (Wegener et al., 2016), as well as none being performed in the paediatric CP population. There appears to be only one paper in English reporting the use of SMotOs on in-toeing gait in children (with idiopathic in-toeing or clubfoot) (Mabuchi et al., 2012). This study by Mabuchi et al. (2012) found that the children improved abnormal gait patterns of paediatric in-toeing gait by decreasing femoral internal rotation through the end of the swing phase and the beginning of the stance phase, and by decreasing tibial internal rotation during the stance phase.

Given the lack of research around SMotO, the aim of this study was to compare the effects of SMotO and AFO on gross motor skills, assessed by GMFM-88 and the BBS in children with CP. It was hypothesised that there would be a significant difference in dynamic gross motor skills as evidenced by a higher GMFM-88 score when wearing SMotO, and static skills when wearing AFOs as evidenced by a higher BBS score.

5.3. Methods

Ethics Process: Ethical approval for the conduct of this study was obtained through the Bond University Research and Ethics Committee (Approval RO-1835). This cross-sectional cohort study investigated the GMFM-88 and BBS in children with CP wearing SMotOs and AFOs.

Setting: Testing was undertaken in 2014 and 2015 in two separate private practice clinic locations (Sydney, NSW, Australia) as well as participant home settings (Sydney, NSW, Australia), accommodating families unable to travel. These settings were familiar to the child, and parents and/or siblings were present throughout the testing.

5.3.1. Participants

5.3.1.1. *Recruitment*

Participants considered for inclusion were children (aged between 2 and 18) with CP recruited via convenience sampling through two private therapy practices (Therapies for Kids and NAPA Centre), Sydney, NSW, Australia.

5.3.1.2. *Eligibility Criteria*

The participant inclusion criteria were: a) a diagnosis of CP, b) no surgery in past six weeks, and c) currently using SMotO and AFOs and having completed the wearing in process of at least two weeks. Participants were excluded if they were unable to follow direction (cognitively).

5.3.2. Intervention / Orthoses

Participants brought their own SMotOs and AFOs to the data collection sessions. The AFOs were previously prescribed through external public or private orthotists and were made from polypropylene with Velcro straps holding the foot in place. The AFOs were either hinged or solid and all had a full-length foot plate. Due to the AFOs being the participants 'usual prescription' there was no further assessment or measurements of the AFOs performed. The prescription of SMotO was assessed by a podiatrist or pedorthists who were both expert in this design type of orthosis and custom made for each child from ethyl vinyl acetate (EVA). Figure 11[5]

[5] Please refer to Figure 11 in Chapter 6 which remains insitu due to paper publication

shows the finished SMotO (Wegener et al., 2016). Figure 12[6] illustrates the placement of toes (circles) metatarsal heads (crosses), and plantar fascia (lines).

5.3.3. Outcome Measures

GMFM-88: This outcome measure is designed specifically for children with CP to evaluate change in gross motor function (Russell et al., 1989). The GMFM-88 is a standardized assessment with demonstrated reliability, validity, and responsiveness to change (Russell & Gorter, 2005).The GMFM-88 was designed to assess 'does do' rather than 'can do'. As such, no assistance is given and an objective definition of items and a standardized scoring system is used (Russell et al., 1989). Items span the spectrum of gross motor activities in five dimensions; lying and rolling, sitting, crawling and kneeling, standing, and walking, running and jumping. The GMFM-88 final score is the amalgamation of the five dimensions of gross motor function. According to the GMFM-88 user manual system, a percent increase of 5-7% of score is classed as a medium positive change (CanChild, 2020).

The BBS is considered to be reliable and valid measure of balance of a child with CP (Iatridou & Dionyssiotis, 2013). The BBS is a 14-section test that covers balance challenges the child may face in day-to-day activity (e.g., static standing without holding on, getting up and down from a chair, picking an object up off the floor). Each section has a five-point rating scale from 0-4, (0 = lowest level of function, 4 = highest level of function). The total score is calculated by adding all section scores, with the highest possible score being 56. The desired score is a higher number as it indicates a reduced risk of falling, whereas the lower number demonstrates a higher risk of falls, therefore reduced safety and balance. The results, in raw scores, are then subject to the following interpretation: 41-56 = low fall risk, 21-40 = medium fall risk, 0 –20 = high fall risk. The minimal clinically important difference (MCID) for the BBS was noted by Gervasoni et al., (2017) as three points of difference in the final score.

5.3.4. Process

Consent was gained from Clinic Directors in both private practice settings. Participants and their caregivers were given an explanatory statement and consent form both of which were read and completed before data collection took place. The outcome measures were conducted by the principal researcher, a paediatric physiotherapist with six years of experience using these tools.

[6] Please refer to Figure 12 in Chapter 6 which remains insitu due to paper publication

The two outcome measures were performed on the same day, in either order, by the participants whilst wearing orthoses, the SMotO and AFO, in either order. During the first measurement, the participant performed both GMFM-88 and BBS in the chosen orthoses. They then had a small break and performed the GMFM-88 and BBS in the other orthoses. The measurements were taken within 30 minutes of initial donning of orthoses and shoes. Due to the participants using their usual orthoses, there did not need to be a 'wear in' or familiarisation timeline.

Statistical Package for Social Science (SPSS) statistics software (Version 20.0, IBM, NY, USA) and the Microsoft Office Excel 2007 (Microsoft, WA, USA) were used for the data entry and analysis. The data for both orthoses was initially analysed for normality, as assessed by skewness and kurtosis values, as well as the use of the Shapiro-Wilk test, and visual inspection of Normal Q-Q Plots and histograms. To determine significance, the data for both orthoses was statistically analysed through a paired t-test if data was normally distributed, or through Wilcoxon signed-rank test if not normally distributed. Descriptive statistics of the total scores (GMFM-88 and the BBS) in each orthosis were used to analyse the data. If the data was normally distributed then the mean and standard difference (SD) was used, if the data was not normally distributed, then the median and interquartile range was use.

5.4. Results

5.4.1. Participants

From 25 possible participants, 10 were unable to participate due to inability to understand instruction or poor comprehension and four were unable to attend data collection. From the 11 potential participants, two were excluded as they did not wear AFOs anymore. See Table 12 for full participant demographics. One participant (Participant 9) was only able to participate in the GMFM-88 as they could not complete any of the BBS tasks safely.

Table 12: Participant demographics

Participant	CP type	GMFCS Level	Age (whole years)	Walking Aid	Outcome Measure
1	Sp Dip	III	3	Reverse walker	GMFM-88 & BBS
2	Sp Q	III	7	Reverse walker	GMFM-88 & BBS
3	Sp Dip	I	4	-	GMFM-88 & BBS
4	Sp Dip	II	4	-	GMFM-88 & BBS
5	Sp Q	IV	3	Rifton Pacer	GMFM-88 & BBS
6	Sp Q	II	6	-	GMFM-88 & BBS
7	Sp Q	II	13	-	GMFM-88 & BBS
8	Sp Dip	III	3	Reverse walker	GMFM-88 & BBS
9	Sp Q	IV	6	Reverse Walker	GMFM-88

BBS = Berg Balance Scale; CP = Cerebral Palsy; GMFCS = Gross Motor Function Classification System; GMFM-88= Gross Motor Function Measure; Sp Dip = Spastic Diplegia; Sp Q = Spastic Quadriplegia

The final yield of participants (n=9; aged between 3 – 13 years with average 5.4 ± 3.2 years), included participants with spastic diplegia (n=4) and spastic quadriplegia (n=5). The nine participants demonstrated a large range of physical ability with reported GMFCS levels of I (n=1), II (n=3), III (n=3) and IV (n=2). Five participants used assistive devices usually but did not require them for this study (refer to Table 14). Participants wore HAFOs (n=4, participants 3, 4, 6 and 7) and the remaining participants wore SAFOs (n=5). One participant (Participant 2) had EVA heel wedges on their SAFO to encourage weight through heel, mimicking heel strike.

5.4.2. The GMFM-88

The individual scores for each of the participants that completed this outcome measure are shown in Table 13 with the statistical results between GMFM-88 scores in AFOs and SMotOs outlined in Table 14. On average, participants scored significantly [t(8)= 4.493, *p*=0.002] higher on the GMFM-88 (mean score difference = 3.59 ± 2.40: 95% CI :1.75, 5.44) when wearing SMotOs compared to AFOs. Overall, the scores demonstrate a trend of higher values for the SMotO group, which is observed through results outlined in Tables 13 and 14.

Table 13: Individual Scores of GMFM-88 (Overall Score/100)

Participant Number	SMotO	AFO	Difference between SMotO and AFO (%)
1	73.51	71.17	+1.98
2	69.11	64.41	+4.70
3	92.31	85.92	+6.39*
4	85.51	79.51	+6.00*
5	46.23	40.43	+5.89*
6	90.75	88.88	+1.87
7	91.29	92.00	-0.71
8	76.97	72.87	+4.30
9	49.52	47.67	+1.85

* = Minimal Clinically Important Difference; GMFM-88 = Gross Motor Function Measure; SMotO = sensomotoric orthoses; AFO = ankle-foot orthoses

Table 14: Mean and SD of total AFO and SMotO GMFM-88 scores

Orthosis	N	Mean	SD
AFO	9	71.43	17.96
SMotO	9	75.02	17.47

SD = Standard Deviation; AFO = ankle-foot orthoses; SMotO = sensomotoric orthoses; GMFM-88 = Gross Motor Function Measure

Three of the participants (n=3 participants 3, 4 and 5) demonstrated medium positive change between orthoses 6.4%, 6.0% and 5.9% respectively. On average participants scored 3.6% mean difference of the GMFM-88 between orthoses when wearing the SMotOs compared to the AFOs.

The total mean and SD of each dimension of the GMFM-88 is displayed in Table 15. The dimensions with a 5-7% increase in score (SMotOs over AFOs) were sections D (6.0%) and E (6.0%) of the GMFM-88.

Table 15: Dimension comparison between orthoses

	AFO		SMOTO		% DIFFERENCE (SMOTO – AFO)
SECTION	**MEAN**	**SD**	**MEAN**	**SD**	
A	98.69	2.19	99.13	1.99	**0.44**
B	89.63	15.45	92.78	12.50	**3.15**
C	78.31	24.58	81.13	23.19	**2.82**
D	54.42	27.25	60.40	27.23	**5.98***
E	36.11	28.48	42.13	32.37	**6.02***

SD = Standard Deviation; AFO = ankle-foot orthoses; SMotO = sensomotoric orthoses; * = Minimal Clinically Important Difference

5.4.3. The BBS

There were eight out of the nine participants who successfully completed the BBS. One was unable due to poor comprehension of task required and poor behaviour. The total mean score for SMotO was 22.00 (±13.00) points and for AFOs was 21.88 (±15.37) points. There was no significant difference between total scores when wearing SMotOs and AFOs [t(7)=.094, *p*=0.928: 95% CI -3.02, 3.27].

The individual BBS scores whilst wearing AFOs and SMotOs are detailed in Table 16. Four out of eight (50%) participants had a three points or higher difference between orthoses indicating MCID. Two of these participants (participants 1 and 4) recorded higher scores in SMotOs and two participants (participants 3 and 6) recorded a higher score in AFOs. The total mean and SD of each dimension of the BBS is displayed in Table 17.

Table 16: Individual participant data from BBS (total score from 56)

Participant Number	SMotO	AFO
1	**15***	12
2	7	7
3	35	**42***
4	**17***	13
5	7	5
6	36	**40***
7	39	37
8	20	19
Total Mean and SD	**22±13**	**21.9±15.4**

* = Minimal Clinically Important Difference; BBS = Berg Balance Scale; SMotO = sensomotoric orthoses; AFO = ankle-foot Orthoses; SD = Standard Deviation

Table 17: Mean and SD for BBS between orthoses

Orthosis	N	Mean	SD
AFO	8	21.88	15.37
SMotO	8	22.00	13.00

BBS = Berg Balance Scale;
SD = standard deviation;
AFO = ankle-foot orthoses;
SMotO = sensomotoric orthoses

5.5. Discussion

The purpose of this study was to investigate the effects of SMotOs compared to AFOs on functional gross motor skills of children with CP through performance of the GMFM-88 and BBS. The hypothesis of a significant difference in gross motor skills as evidenced by a higher GMFM-88 score when wearing SMotO, and as evidenced by a higher BBS score when wearing AFOs was confirmed.

An increased score of 5-7% was demonstrated in three participants (6.4%, 6.0% and 5.9%) when wearing SMotOs over AFOs, which, as per the GMFM-88 is a medium positive change. Looking at the total mean for each dimension, it appears that the walking and running segments (dimensions D and E) demonstrated the largest difference in scores between AFO and SMotO. The SMotOs had the higher score, which would infer the skills were easier to perform, in turn aligning with the theory that the SMotOs allow for more dynamic movements than AFOs. As SMotOs are more dynamic than AFOs, the results somewhat support the studies by Dalvand et al. (2013) and Zhang et al. (2009) who reported HAFOs demonstrated higher improvements in GMFM than when in SAFOs. As this study found children in AFOs had poorer results than SMotOs, it could be suggested that SMotOs could be an even better option for orthosis when searching for improved dynamic function.

The total GMFM-88 scores showed there was a significant (p=0.002) difference between GMFM-88 scores when wearing SMotOs over AFOs. These results suggested that children with CP who wear SMotOs had a significantly higher level of gross motor skills than when they wear AFOs. This may be likely due to the dynamic nature of the SMotOs compared to the larger, more awkward and restrictive nature of AFOs. As a deficit in gross motor skills are a hallmark indicator for severity of CP, improving gross motor skills is key to improving function, therefore the possibility of reducing the severity of the appearance of their CP.

The BBS was a little more complex to undertake than the GMFM-88 as there were specific instructions to follow, with a higher level of skill needed from the participants. There were four participants who demonstrated improvements in balance between orthoses. Two (Participants 1 and 4) out of the eight participants demonstrated an MCID of three points of difference or more when wearing SMotOs, and two (Participant 3 and 6) out of the eight participants who demonstrated an MCID of three points of difference or more when wearing AFOs. There were no definitive differences in the types or levels of CP in those who made significant improvements in balance. Overall, the total mean score of each orthoses in the BBS did not show any significant differences. This contradicts the findings by Zhang et al. (2009) who found improvements in BBS when wearing HAFOs compared to SAFOs. As there were a small participant group for the BBS, the results require further investigation into comparing the effects of the SMotO and AFO on the BBS.

5.5.1. Limitations

Low numbers with recruitment was a limitation of this study. This was due to the complex nature of the clinic whereby participants may have been prescribed orthoses but had not yet completed the wearing in time before leaving the clinic. Furthermore, the high level of complex diagnoses in the clinics used meant there were a notable number of children excluded due to not meeting the inclusion criteria for diagnosis. The study design also presented as a limitation. To properly compare the effect of the orthoses, including barefoot comparisons would have demonstrated a baseline score of each participant. Including the extra comparison was not feasible for this population as completing six rounds of outcome measures would have been too tiring, both mentally and physically. Noting this study design limitation, the approach taken in this study did allow for the ability to capture data that was less likely to be confounded by fatigue. Another limitation is not investigating the long-term effect of the orthoses on the OMs, but this could be included in future studies.

5.5.2. Clinical application

The GMFM-88 was found to be relatively easy to employ to assess the participants as some of the tasks were able to be completed without verbal, receptive or expressive language. Rather, the participant was cued through motivation to reach for a toy or was placed in a position to assess movement out of the position. Many of the families appeared comfortable with the application and sequence of this outcome measure as it had been seen in assessment sessions conducted in the past.

The BBS appeared to be a little more complex to employ as participants had to follow specific directions. It may be more clinically relevant to use alternate balance scale measures when assessing children with cognition difficulties. When observing parents assisting the participants don and doff the orthoses, the AFOs appeared to require more time and effort to apply, whereas the SMotOs appeared to be donned faster. It may be that there is an increased ease of use with the SMotOs, but this would require further investigation.

5.6. Conclusion

The results demonstrated a preliminary insight and initial support of an effective benefit on gross motor skills when wearing SMotOs compared to when wearing AFOs, when assessed using the GMFM-88. This finding infers SMotOs may be a viable alternate or supplementary orthotic option for children with CP; however, further investigation is required. SMotOs may also provide an increased ease of use due to their low-profile design compared to AFOs.

6.0. Using the Edinburgh Visual Gait Score to Compare Ankle-Foot Orthoses, Sensorimotor Orthoses and Barefoot Gait Pattern in Children with Cerebral Palsy

Prelude

The following chapter continues the theme of assessing the effect of sensomotoric orthoses and ankle-foot orthoses in children with cerebral palsy but with the focus on gait quality. Gait was assessed using the outcome measure defined in Chapter 4 (Appendix D, Section iv). The study reported in this chapter was published in the open access journal Children*. Please find the full PDF version in Appendix A.*

6.1. Abstract

Gait analysis is one aspect of evaluation in ambulatory children with cerebral palsy (CP). Ankle-foot orthoses (AFOs) improve gait and alignment through providing support. An alternative and under-researched orthosis are sensomotoric orthoses (SMotOs). The Edinburgh Visual Gait Score (EVGS) is a valid observational gait analysis scale to measure gait quality. The aim of this study was to use the EVGS to determine what effect AFOs and SMotOs have on gait in children with CP. The inclusion criteria were: a) mobilising children with CP diagnosis, b) no surgery in past six weeks and c) currently using SMotOs and AFOs. Eleven participants were videoed walking 5m (any order) barefoot, in SMotOs and AFOs. Of the participants (age range 3 – 13 years, mean 5.5 ± 2.9) two were female and six used assistive devices. Seven could walk barefoot. Participants had spastic diplegia (n=4), spastic quadriplegia (n=6) and spastic dystonic quadriplegia (n=1). Gross Motor Functional Classification System (GMFCS) levels ranged I - IV. Total score for SMotOs (7.62) and AFOs (14.18) demonstrated improved gait when wearing SMotOs (no significant differences between barefoot and AFOs). SMotOs may be a viable option to improve gait in this population. Additional study is required but SMotOs may be useful in clinical settings.

6.2. Introduction

Cerebral Palsy (CP) is a neurodevelopmental condition well recognised to begin at birth or early childhood and persisting through the lifespan (Rosenbaum, Paneth, Leviton, Goldsteing and Bax, 2007). It has been defined as a group of permanent disorders of the development of movement and postures, causing activity limitation through spasticity (Romkes & Brunner, 2002; Wingstrand, Hägglund, & Rodby-Bousquet, 2014), muscle weakness, impaired postural control, and selective motor control as some of the primary manifestations of this brain injury (Wingstrand et al., 2014). It is often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, and by epilepsy. One of these activity limitations may be the ability and co-ordination for walking (gait), with control of movements and postures being affected. As ambulation is the usual method for mobilising, many children with CP strive to achieve any form of walking possible, whether it's with or without an assistive device.

Gait assessment assists in determining the degree and cause of gait abnormality and can be used as an outcome measure to evaluate change and the effectiveness of intervention (Harvey & Gorter, 2011; Kawamura et al., 2007; Rathinam, Bateman, Peirson & Skinner, 2014). Instrumented gait analysis is the gold standard for evaluation of movement (Rathinam et al., 2014) but requires highly technological equipment in a specialised gait laboratory. A gait laboratory requires considerable capital investment, trained personnel, and is not often readily accessible for routine clinical work (Dickens, 2006; Harvey & Gorter, 2011). Observational gait assessment is considered as a cost-effective alternate for instrumented gait analysis in regular clinical practice (Rathinam et al., 2014). One observational tool used in clinical settings is the Edinburgh Visual Gait Score (EVGS). The EVGS has been demonstrated as a valid and reliable (del Pilar Duque Orozco et al., 2016; Read, Hazlewood, Hillman, Prescott, & Robb, 2003), clinically applicable visual gait analyses tool for children with CP (Read et al., 2003).

Ankle-foot orthotics (AFOs) are the typical prescription of lower extremity orthoses for the management of lower limb deformities that often occur with CP. A consensus conference of the International Society for Prosthetics and Orthotics identified the aims of lower extremity orthotic management in children with CP as to 1) correct and/or prevent deformity, 2) provide a base of support, 3) facilitate training of motor skills, and 4) improve efficiency of walking (Morris & Condie, 2009). The goal of each AFO prescribed for a child with CP is the collective improvement of these biomechanical variables to increase the ease of taking an individual step with the potential to enhance walking activity and functional skills (Bjornson et al., 2016). AFOs are designed to: affect body structure (Wingstrand et al., 2014), support normal joint alignment and mechanics, provide variable range of motion (ROM) when appropriate, facilitate function (Brodke et al., 1989; Knutson & Clark, 1991; White, Jenkins, Neace, Tylkowski, & Walker, 2002; Wingstrand et al., 2014;), stabilize the ankle / foot complex (Buckon, Jakobson-Huston, Moor, Sussman, & Aiona, 2004) and enable a continuous Achilles / gastrocnemius stretch (Boyd, Pliatsios, Starr, Wolfe, & Graham, 2000; Hainsworth, Harrison, Sheldon, & Roussounis, 2007; Morris, 2002).

Along with joint alignment, other improvements that may be seen through the use of AFOs are the improvement in walking efficiency (Morris & Condie, 2009; Rethlefsen, Kay, Dennis, Forstein, & Tolo, 1999), position the foot for function (Kane, Musselman, Manns, & Lanovaz, 2016), and improvement in gait function and pain prevention (Westberry et al., 2007).

There are multiple studies assessing and comparing the motor changes in gait and other gross motor skills using different types of AFOs in children with CP (Buckon et al., 2001, 2004; Dalvand, Dehghan, Feizi, Hosseini, & Armirsalari, 2013). Buckon et al. (2004), noted that the AFOs did not significantly improve standing skills of the GMFM-88 ($p \leq 0.025$), but reported an improvement of the walking, running and jumping elements when wearing AFOs ($p < 0.002$) compared to barefoot. In another study, Buckon et al. (2001) noted that despite the AFO enhancing stability throughout static (e.g., standing) and dynamic (e.g., walking) gross motor and functional skills, they did not allow the child to achieve a skill they were previously unable to master. Dalvand et al. (2013) also noted improvements in gait with both solid-AFO (SAFO) and hinged-AFO (HAFO), although HAFO demonstrated better improvements in gross motor function than SAFO.

Unlike AFOs, sensomotoric orthotics (SMotO) provide a different approach to the management of gait in children with CP. Wegener, Wegener, Smith, Schott, & Burns (2016) describe one adaptation theory as ‘elements’ on the foot orthoses (e.g., forefoot valgus posting or lateral rearfoot padding) increase local pressures which are detected by cutaneous receptors, muscle spindles or Golgi apparatus. To expand this, SMotOs are created to directly change the muscle length (Subke, Kolling, Griesemann, Kleinau, & Staudt, 2009) and ‘activate and deactivate’ muscles by increasing or decreasing individually placed point specific pressure on musculotendinous structures in the foot of the tibialis posterior, peroneus brevis and the lumbricals/quadratus plantae. This theory implies the information that is transmitted by the sensors for the control of muscle activity is changed (Ohlendorf, 2013). Depending on these individual pressure bumps’ height and placement, the muscles can be activated or restricted (Ohlendorf, 2013; Subke et al., 2009). CP affects the different areas of the brain, therefore interrupting signals sent to the muscles. The SMotOs work on the idea that the signals are being sent from the muscles back up to the spinal cord through activation of the Golgi bodies, therefore signalling muscles to respond to stimulus.

Clinically, the SMotO have demonstrated improved foot alignment, balance, control with walking and functional skills. Clients have been prescribed the SMotOs as a supplementary lower limb orthosis when it is noted they are finding functional movement restricted in the AFO. SMotOs have also been prescribed for children who require more feedback from their feet, where wearing shoes alone has not been effective.

There is a lack of evidence examining the ‘sensorimotor response’ paradigm, as there are no randomized trials, minimal peer-reviewed papers in English and only a few small cross-sectional paediatric papers (Wegener et al., 2016). There was only one paper in English found that reported the use of SMotOs on in-toeing gait in children (with idiopathic in-toeing or clubfoot). This study found SMotOs improved abnormal gait patterns of paediatric in-toeing gait by decreasing femoral internal rotation through the end of the swing phase and the beginning of the stance phase and by decreasing tibial internal rotation during the stance phase (Mabuchi et al., 2012).

There are numerous papers that demonstrate improved gait when wearing AFOS, but there have been no studies to date comparing the effect of SMotOs and AFOs on gait in children with CP. Clinically, there appears to be improvements in gait quality when children with CP wear SMotOs. The hypothesis was that there would be a better EVGS result when children with CP wore the SMotOs compared to AFOs. Therefore, this study aims to compare the changes in gait from barefoot when children with CP are wearing SMotOs and AFOs, through use of the EVGS.

6.3. Methods

Ethical approval was obtained through the Bond University Research and Ethics Committee (Approval RO-1835). Consent was gained from Clinic Directors in both private practice settings. Participants and their caregivers were given an explanatory statement and consent form both of which were read and completed before data collection took place.

6.3.1. Participants

The participant inclusion criteria were: a) child with a diagnosis of CP, b) no surgery in past six weeks, c) currently using SMotO and AFOs (completed the wearing in process of at least two weeks) and, d) able to mobilize (with or without a device). Participants were recruited by convenience sampling and were assessed through two private paediatric therapy practices in Sydney (Therapies for Kids and NAPA Centre). There was no limit of participation due to GMFCS level. Participants brought their own SMotOs and AFOs to the data collection sessions.

6.3.2. Orthoses

The AFOs were custom-made through public or private orthotists from polypropylene with Velcro straps holding the foot in place. The AFOs all had a full-length foot plate. Due to the AFOs being the participants ‘usual prescription’ there was no further assessment or measurements of the AFOs performed. The SMotOs were custom made for each child from ethyl vinyl acetate (EVA) and had been assessed and prescribed by a podiatrist or pedorthist who were both expert in this design type of orthosis. Figure 11 shows the finished SMotO (Wegener et al., 2016). Figure 12 illustrates the placement of toes (circles) metatarsal heads (crosses), and plantar fascia (lines).

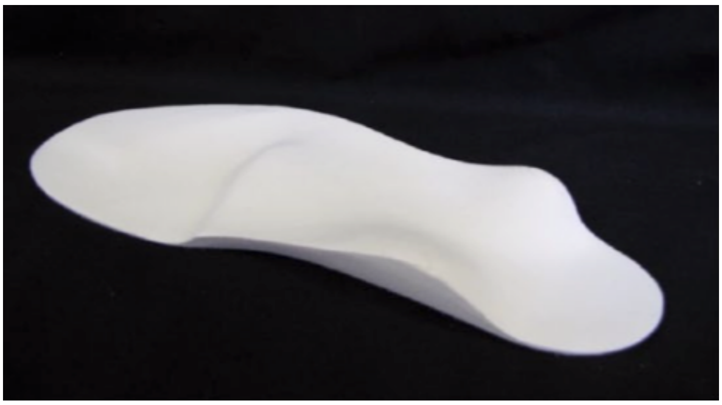


Figure 11: Sensorimotor Orthosis (Wegener et al., 2016)

Figure 12: Sensorimotor Orthosis with descriptive markings

The Golgi bodies (in the musculotendinous junctions of the tendons of the foot) are activated to switch the muscle 'on' or 'off' by the pressure from the 'bumps' coinciding with the musculotendinous junctions (Jarling, 2001) (Figures 11 and 12). It has been proposed that information is sent by afferent feedback pathways (centrally) in order to reduce the activity of over-active muscles through inhibition, which in turn facilitates an increase in the activity of weaker muscles (Ludwig, Quadflieg, & Koch, 2013). Given the physiology of ascending muscle chains, the reaction will not only affect the single muscle targeted (e.g., in the foot) but influence the complete chain of movement (e.g., ankle and lower limb) and positively impact malposition (e.g., over pronation) (Ohlendorf, 2013).

The AFOs and SMotOs were worn inside the participants usual shoe (for that particular orthoses). Participants were asked to walk barefoot (where appropriate, due to ability to mobilize without any type of foot support), in SMotOs and in AFOs for at least 5m at a self-directed pace. To provide motivation, participants were able to choose the order in which they wore the orthoses. Video imagery was taken with a handheld device (Apple iPhone 7s, Apple Inc., CA, USA) in anterior, posterior and lateral views. Videos were taken in a well-lit environment, following for lateral views or zooming in and out (as needed) when the child walked. If required due to poor video quality, misbehaviour or misstep, additional videos were taken to ensure a quality video was assessed. The child was allowed the comfort of a break between walks if needed. To ensure validity and reliability of video, the plane of motion was followed.

6.3.3. Outcome Measure

To determine changes in gait, the EVGS was used. The EVGS comprises 17 parameters for each lower limb and evaluates movement across six sites (trunk, pelvis, hip, knee, ankle and foot) (Read et al., 2003). Each gait phase is analysed in the frontal, sagittal and transverse planes and the anatomical sites are evaluated for movement through video observation (Bella, Rodrigues, Valenciano, Silva, & Souza, 2012). Scoring uses a 3-point ordinal scale. When the segment is marked 0, it determines a normal score. When there is a 1, it means a moderate deviation from normal in either direction, and 2 relates to a marked deviation, therefore a higher score relates to a more severe deviation or abnormality of gait. The developers of EVGS reported a score reduction of 4 on each limb (compared to pre-intervention score) as an improvement and as the minimum change in score required that would be indicative of change, not merely related to observer variation (Read et al., 2003).

The EVGS is a valid, robust, reliable and easy-to-use observational gait analysis scale to measure of gait quality in children with CP (Rathinam et al., 2014; Read et al., 2003; Robinson, Clement, Herman, & Gaston, 2017). It has been examined for the purpose of orthosis evaluation in adults (Putnam, Wening, & Hasso, 2014) but not yet validated in children with orthoses. The scale has stringent instructions to ensure reliability. Its agreement and validity with three-dimensional gait analysis has been documented (del Pilar Duque Orozco et al., 2016) and was noted to be 52-73%. The essential properties of an observation scale are validity, reliability, and ability to detect change (Gupta & Raja, 2012). Responsiveness is the ability of a tool's detection of change due to an intervention or over time. The EVGS is shown to correlate with the Gait Profile Score and the GMFCS (Robinson et al., 2015) two relevant and valid measures relating to CP. Frame by frame analysis was performed to score the gait using the EVGS with all analysis performed by the principal researcher, a physiotherapist who works with the children and has 6 years of experience using this tool. This analysis took place after all face-to-face data collection had been completed thereby minimizing time pressures on the families and their child. As the participant walked 5m for each camera direction, there was more than one stride to observe. Once viewed, the most usual score was used.

6.3.4. Data Analysis

The EVGS was analysed through SPSS statistics software (Version 20.0, IBM, NY, USA) and the Microsoft Office Excel 2007 (Microsoft, WA, USA) were used for the data entry and analysis. Normality was determined via visual inspection of histograms, box plots and normal Q-Q plots. Depending on distribution, parametric or non-parametric tests were used to determine if there were any significant differences in the baseline characteristics or of the groups. Descriptive statistics were used to profile the data: the median difference of the total EVGS scores and the mean difference of the average walking score, before and after the intervention, were calculated. A summated score of each limb was used for data analysis for this study. Thus, the score for the EVGS ranges from 0 to 34 on left (L) or right (R). The data for the barefoot condition and both orthoses was then statistically analysed through a one-way analysis of variance (ANOVA) to determine significance and post hoc Bonferroni to outline further comparison significance. There was also a cumulative total of each segment analysed. Repeated measures ANOVA with Bonferroni post hoc and Wilk's Λ was performed. Alpha levels were set at 0.05 *a priori.*

6.4. Results

6.4.1. Participants

From 27 possible participants, 10 were unable to participate due to inability to understand instruction or poor comprehension and four were unable to attend data collection. From the 13 potential participants, two were excluded as they did not wear AFOs anymore. See Table 18 for full participant demographics.

Table 18: Participant demographics

Participant	CP type	GMFCS Level	Age (whole years)	Walking Aid
1	Sp Dip	III	3	Reverse walker
2	Sp Q	III	7	Reverse walker
3	Sp Dip	I	4	-
4	Sp Dip	II	4	-
5	Sp Q	IV	3	Rifton Pacer
6	Sp Q	II	6	-
7	Sp Q	II	13	-
8	Sp Dip	III	3	Reverse walker
9	Sp Dys Q	IV	4	Buddy Roamer
10	Sp Q	IV	6	Reverse walker
11	Sp Q	II	8	-

CP=Cerebral Palsy; Sp Dip=Spastic Diplegia; Sp Q=Spastic Quadriplegia; Sp Dys Q=Spastic Dystonic Quadriplegia; CP=Cerebral Palsy

Of the final yield of 11 participants (aged between 3 – 13 years with average 5.5 ± 2.9 years), seven were able to walk barefoot and therefore had barefoot data collection recorded. Four were unable to walk barefoot due to inability or child refusal. The participants had spastic diplegia (n=4), spastic quadriplegia (n=6) and spastic dystonic quadriplegia (n=1). The GMFCS levels of participants were: level I (n=1), level II (n=4), level III (n=3), level IV (n=3). Six participants used assistive devices for EVGS (Participants 1, 2, 8 and 11 used a reverse walker, Participant 6 used a Rifton pacer, Participant 10 used a Buddy Roamer). Five participants wore HAFOs (Participants 3, 4, 6, 7 and 11) and the remaining participants wore SAFOs. One participant had EVA heel wedges on their SAFO to encourage weight through heel, mimicking heel strike. There was no data recorded of orthosis use timing per child over usual day due to this research focusing on the immediate effect of orthoses on gait.

6.4.2. Scores

The total EVGS for (L) and (R) barefoot (where applicable) and with each orthosis are described in Table 19.

Table 19: Total (L) and (R) EVGS for participants

Participant	GMFCS Level	Total score barefoot		Total score AFO		Total score SMotO	
		(L)	(R)	(L)	(R)	(L)	(R)
1	III	26	27	20	21	14	16
2	III	11	13	6	9	4	4
3	I	20	16	12	12	17	10
4	II	11	15	16	15	7	8
5	IV	13	9	12	11	8	8
6	II	25	26	14	16	7	10
7	II	12	14	8	9	4	5
8	III	- [f]	- [f]	18	20	11	14
9	IV	- [f]	- [f]	20	20	12	12
10	IV	- [f]	- [f]	14	14	6	6
11	II	- [f]	- [f]	14	11	4	5

GMFCS=Gross Motor Functional Classification Scale; (L)=left; (R)=right; AFO=ankle-foot orthoses; SMotO=sensomotoric orthoses; [f] No data due to inability or refusal to walk barefoot.

When barefoot was assessed, the overall scores across participants demonstrated a poorer score than both AFOs and SMotOs except for one participant, who demonstrated poorer results when wearing AFOs compared to barefoot and SMotOs. The descriptive statistics of EVGS is provided in Table 20 outlining the mean and SD for total (L) and (R) scores for barefoot, AFO and SMotO intervention.

Table 20: Data descriptors for EVGS total (L) and (R)

Intervention		N	Mean	Std. Deviation
EVGS Total (L)	Barefoot	7	16.86	6.67
	AFO	11	14.00	4.47
	SMotO	11	8.55	4.43
Total (L)		**29**	**12.62**	**5.94**
EVGS Total (R)	Barefoot	7	17.14	6.77
	AFO	11	14.36	4.43
	SMotO	11	8.91	3.91
Total (R)		**29**	**12.97**	**5.82**

EVGS=Edinburgh Visual Gait Score; (L)=left; AFO=ankle-foot orthoses; SMotO=sensomotoric orthoses; (R)=right.

One-way ANOVA analyses revealed significant differences between total (L) (p=0.011) and (R) (p=0.014) scores between SMotO and AFOs (Table 21).

Table 21: Bonferroni comparison between the three conditions

	Intervention	Intervention	Significance
EVGS Total (L)	Barefoot	AFO	1.0
		SMotO	0.032*
	AFO	Barefoot	1.0
		SMotO	0.027*
EVGS Total (R)	Barefoot	AFO	1.0
		SMotO	0.052
	AFO	Barefoot	1.0
		SMotO	0.028*

EVGS=Edinburgh Visual Gait Score; (L)=left; AFO=ankle-foot orthoses; SMotO=sensomotoric orthoses; (R)=right; * Indicates significance differences.

There were significant differences on the (L) lower limb between barefoot and SMotO (p=0.032), and AFO and SMotO (p=0.027). On the (R) lower limb, there was a significant difference between AFO and SMotO (p=0.028).

In the segmental analyses, repeated measures ANOVA elicited statistically significant differences in the foot, $F(2, 5)=8.993$, $p<0.022$; Wilk's $\Lambda=0.218$, partial $\eta2=0.782$, and hip, $F(2,5)=6.10$, $p<0.045$; Wilk's $\Lambda=0.290$, partial $\eta2=0.710$, with the biggest effect in the foot. Post hoc analysis with a Bonferroni adjustment revealed statistically significant differences between barefoot and SMotOs in the foot, mean difference (MD)=8.86 (95% confidence interval [CI] 2.38 to 15.33, p=0.012), and between AFO and SMotOs at the hip, MD=1.14 (95% CI, 0.03 to 2.26, p=0.046).

6.5. Discussion

The aim of this study was to investigate the changes two types of orthoses (SMotOs and AFOs) had on gait pattern in children with CP as derived through the EVGS. The hypothesis that there would be a better EVGS result when children with CP wore the SMotOs compared to AFOs was confirmed. All the participants were in GMFCS levels I-IV and used orthoses to walk, both in SMotOs and AFOs. There were six participants who required the use ambulatory aids, displaying a varied range of gait ability. Overall, this cross-sectional cohort study found SMotOs to have a more positive influence on gait pattern compared to AFOs and barefoot.

The total raw scores of each participant demonstrated more desirable gait patterns were observed when wearing SMotO. This was resultant across 11 participants by a lower total score when wearing SMotOs on both left (7.46) and right

(7.77) compared to when wearing AFOs on left (14.00) and right (14.36) and the seven participants with barefoot left (14.22) and right (14.11). Due to limitations in assessing calcaneal alignment in orthoses, the EVGS results would be affected at the foot / ankle when using a restrictive orthoses compared to a more dynamic orthoses that does not limit ankle movement. Barring participants 3 and 5, the general trend indicated that SMotO had the lower scores, which correlates to the EVGS score line indicating a more aligned gait pattern. The one-way ANOVA confirmed that there was a significant difference in EVGS scores between the use of SMotO and AFO. One participant demonstrated a worse score on the (L) foot when wearing SMotO compared to AFO due to poor foot and knee alignment. Subsequently, a significant difference on the left lower limb was found when participants wore SMotOs compared to barefoot or AFOs, but not the right lower limb. The right lower extremity score was close to being significant (p=0.052) when comparing SMotO to barefoot or AFOs but may need a larger yield study to determine its significance and if it is a usual trend. Interestingly, there was no significant difference between the AFO and barefoot scores.

Looking at the segmental breakdown of the EVGS, the hip and foot were seen to be most affected by orthotic intervention. It was found that that in the foot, barefoot is significantly different from SMotO (p=0.012) and in the hip there is a significant difference between the AFO and SMotO (p=0.046). The differences in the foot results between barefoot and SMotO may demonstrate the theory presented earlier by Wegner et al. (2016). Interestingly, there was no difference noted at the pelvis between barefoot and AFO. At the trunk, the AFOs presented a higher score than either barefoot or SMotOs, but this was not significant.

With regard to using the EVGS as an outcome measure to assess the effect of orthoses, a search for papers assessing the effect of AFOs on gait in children with CP through the EVGS only resulted in one case study paper by Young and Jackson (2019). This paper followed a child with spastic bilateral CP over 15-months, in which she was prescribed AFOs and began to stand and walk independently. Post AFO prescription, they noted minimal clinically important differences (MCID) in EVGS (increase by 7 points on the left and 11 points on the right – MCID of 2.4) and gait speed (42.9% increase in speed – MCID >10.9% is noted as large). This paper noted that AFOs did create a significant improve in gait compared to barefoot. The lack of multiple papers using the EVGS to compare the effects of AFOs on gait in children with CP may provide a direction for future research.

Some of the most common gait anomalies found in children with CP was in-toeing (amongst others) (Rethlefsen et al., 2017). Although in a different population, there was one paper supporting the use of SMotOs to correct in-toeing in children with idiopathic in-toeing gait or clubfoot. Mabuchi et al. (2012) assessed the biomechanical effect of these orthoses on in-toeing gait in children. They found that the orthoses showed significant decreased in internal rotation at the proximal femur (loading response phase $-18.3^{\circ}\pm28.1^{\circ}$ versus $-21.6^{\circ}\pm28.0^{\circ}$, $p=0.009$ and terminal swing phase $-16.3^{\circ}\pm27.4^{\circ}$ versus $-19.0^{\circ}\pm26.4^{\circ}$, $p=0.047$) and the tibia in mid stance phase ($0.7^{\circ}\pm 12.5^{\circ}$ versus $-2.0^{\circ}\pm 14.9^{\circ}$, $p=0.030$) and terminal stance phase ($1.4^{\circ}\pm11.9^{\circ}$ versus $-2.3^{\circ}\pm14.5^{\circ}$, $p=0.042$). They also found a significant increase in walking speed (67.9 m/min versus 64.9 m/min, $p<0.001$) and stride length (500 mm versus 477 mm, $p<0.001$). This may provide a basis to address in-toeing in children with CP with SMotOs.

The small, heterogenous sample affects the strength and generalizability of the results. Therefore, it is recommended that future research includes larger, more homogenous samples investigating SMotOs as another form of orthotic therapy for children with varied types of CP. It may also create opportunities to further investigate clinically useful observational gait assessment tools, such as the EVGS, for outcome measures when prescribing interventions such as orthoses. In support of Jagademma et al. (2015), who stated that when investigating the effects of interventions such as AFOs, it is important to categorise children with CP based on their gait abnormalities. Therefore, it may be beneficial to further investigate multiple orthoses options or combinations than just AFOs alone, depending on the child's needs. Future research could include validating the EVGS as clinical assessment tool for use in children with lower limb orthoses, comparing customised, tuned AFOs with SMotOs in three-dimensional gait analysis, or expanding this study by removing limitations and performing over a longer time period.

6.5.1. Limitations

Due to the participants being recruited from a specific population, there were limitations on the number of potential participants. A study on 11 variable patients using both assistive devices does not provide evidence of superiority of either orthoses. For example, a patient may have better foot kinematics with the SMotO in isolation but may collapse at the knee. Limitations to this research include a final small yield of participants available to collect the full range of data, mainly due to the restrictions uncovered such as comprehension of task, ability to follow direction, and ability to attend data collection. This may affect the strength of the results, although papers within the literature demonstrate a range of participant numbers whilst using the EVGS from 7 (Bella et al., 2012) to 151 (Robinson et al., 2015). With a longer recruitment period and implementation of this as 'usual clinical practice' in prescription of SMotOs, a larger group could be assessed for continuation of these research findings. Another limitation was the lack of tuning the AFOs to avoid the possibility of iatrogenic gait compensations. Tuning of AFOs is recommended by Owen,(2010) and Eddison & Chockalingam (2013) but was not performed in this setting as the researchers were investigating the usual prescription of AFOs and SMotOs on gait pattern. The ankle, nor ankle in AFOs, were assessed for ankle range of motion during this study which may provide limitation to the strength of this study. Ideally, this study would have provided customised AFOs with in-depth explanation of prescription process. This would thus enhance the quality of study and reduce possible suboptimal AFO prescription. Unfortunately, this process is complex, can be expensive and was not possible at this stage. Potential bias is another limitation acknowledged in this paper and would be better resolved with EVGS completed by blinded raters with experience as well as more stringent patient preparation and video capture methodology. Along with this, the EVGS has not been assessed for reliability and validity when observing children in orthoses, possibly due to the inability to observe the calcaneus in orthoses or shoes. Ong, Hillman & Robb (2008) validated the EVGS' reliability and validity for experienced observers in gait analysis. They noted however, that the inexperienced observers were less accurate, and the experienced observers demonstrated more accurate results when compared to three-dimensional gait analysis. Three-dimensional gait analysis would be the preferred method of assessment for this type of study, but access to such a system was not possible. The EVGS does not allow for specific reporting of deviation direction, but rather indicated a deviation from 'normal', which may not be enough detail for some gait analyses.

6.6. Conclusion

The results of this study suggest that further evaluation of the effects of SMotO are warranted but the SMotO may, clinically, be an effective orthosis intervention to improve gait in children with CP. These results may be better validated if further research is performed in a gait laboratory using the gold standard three-dimensional gait analysis versus an observational score as there are many aspects of gait analysis. These results encourage further investigation into the use of SMotO in children with CP or to further specify the areas of benefit of the SMotO alongside AFO in relation to this population, their gait function and level of disability. Clinically, this creates an alternate orthoses prescription possibility for children with CP.

7.0. Qualitative Analysis of Sensomotoric Orthoses & Ankle-Foot Orthoses in Children with Cerebral Palsy: A Quality-of-Life Questionnaire

Prelude

This chapter presents a study that was designed to further investigate the effect of sensomotoric orthoses and ankle-foot orthoses on the quality of life in children with cerebral palsy and their families. The Cerebral Palsy Quality of Life questionnaire was deemed inappropriate (as per Study 4, Chapter 4). As there were no questionnaires surveying these particular subjects, a specific questionnaire was designed to help further determine quality of life themes and further inform the effect of sensomotoric orthoses and ankle-foot orthoses on quality of life in children with cerebral palsy.

7.1. Introduction

Cerebral palsy (CP) is a term used to describe a group of permanent, non-progressive, disorders affecting the development of human movement and postures. The condition is caused early in life primarily by a brain injury or lesion (Danino et al., 2015) and leads to physical activity limitations in the locomotor apparatus (Christovao et al., 2015; Ridgewell, Dobson, Bach, & Baker, 2010; Rosenbaum, Paneth, Leviton, Goldstein, & Bax, 2007; Romkes & Brunner, 2002). It causes activity limitation through spasticity (Romkes & Brunner, 2002; Wingstrand, Hägglund, & Rodby-Bousquet, 2014), muscle weakness, impaired postural control, and selective motor control as some of the primary manifestations of this brain injury (Wingstrand et al., 2014). In children with CP the leading manifestation of the condition is motor impairments, with subsequent alterations in the body's biomechanical movements (Christovao et al., 2015). Following diagnosis (usually identified in early childhood) this population is commonly treated with physiotherapy, botulinum A toxin injections, and lower extremity orthoses (Morris, 2002).

Quality of life (QoL) is defined by the World Health Organisation (2006) as 'health is a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity. The enjoyment of the highest attainable standard of health is one of the fundamental rights of every human being without distinction of race, religion, political belief, economic or social condition.' With regard to CP, factors relating to QoL can include: child (age, gender, and severity of the disease; comorbidity and complications), family (socioeconomic status, relationships and support, coping mechanisms, parenting style, and knowledge about the disease) and availability of management and rehabilitation services and other environmental factors (Mohammed, Ali, & Mustafa, 2016). In a QoL study by Dickinson et al. (2007), children with CP were investigated using KIDSCREEN (2006) and found, through a comparison of least and most able groups, severely limited self-mobility was significantly associated with reduced mean score for physical wellbeing (7.6, 95% CI 2.7–12.4, p=0.002); and pain was common and associated with lower QoL on all domains. They concluded that physical impairments and presence of pain were responsible for variation (3% and 7%, respectively) in QoL, therefore a child's pain should be carefully assessed. When physical impairment impact function, thus affecting QoL, therapists would likely make improving function a goal area for therapy.

Independent walking is a typical goal of rehabilitation in children with CP, but this expectation can lead to frustration in parents and children, many of whom feel that they are more mobile and more functional when using assistive devices (Gibson, 2013) versus completely independent.

Lower extremity orthoses are often used to address the biomechanical limitations associated with CP and optimise joint alignment (Bjornson, Schmale, Adamczyk-Foster, & McLaughlin, 2006). A consensus conference of the International Society for Prosthetics and Orthotics identified the aims of lower extremity orthotic management in children with CP to 1) correct and/or prevent deformity, 2) provide a base of support, 3) facilitate training of motor skills, and 4) improve efficiency of walking (Morris & Condie, 2009). In children with CP, these orthoses are also used to promote functional activities (Figueiredo, Ferreira, Moreira, Kirkwood, & Fetters, 2008).

The typical prescription of lower extremity orthoses is the ankle foot orthosis (AFOs) (Ries, 2017). The goal of each AFO prescribed for a child with CP is the collective improvement of biomechanical variables with the potential to enhance walking activity and functional skills (Bjornson et al., 2016). AFOs are designed to: affect body structure, support normal joint alignment and mechanics, provide variable range of motion (ROM) when appropriate, prevent pain and facilitate function (Boyd, Pliatsios, Starr, Wolfe, & Graham, 2000; Brodke et al., 1989; Hainsworth, Harrison, Sheldon, & Roussounis, 2007; Kane, Musselman, Manns, & Lanovaz, 2016; Knutson & Clark, 1991; Westberry et al., 2007; White, Jenkins, Neace, Tylkowski, & Walker, 2002; Wingstrand et al., 2014). Common styles of AFOs, as reported by Wren, et al. (2015), include solid-AFOs (SAFO) and hinged-AFOs (HAFO) with dorsiflexion (DF) or plantarflexion (PF) stops, and dynamic-AFOs (DAFO).

Unlike AFOs, sensomotoric orthotics (SMotO) provide a different approach to the management of gait and spasticity in children with CP. Wegener, Wegener, Smith, Schott and Burns (2016) describe one adaptation theory as ‘elements’ on the foot orthoses (e.g. forefoot valgus posting or lateral rearfoot padding) designed to directly change the muscle length (Subke, Kolling, Griesemann, Kleinau, & Staudt, 2009) by increasing local pressures which are detected by cutaneous receptors, muscle spindles or Golgi apparatus (Ohlendorf, 2013). It has been proposed that information is sent by afferent feedback pathways in order to reduce the activity of over-active muscles through inhibition, which in turn facilitates an increase in the activity of weaker muscles (Ludwig, Quadflieg, & Koch, 2013; Ohlendorf, 2013). Specifically, musculotendinous structures in the foot of the tibialis posterior, peroneus brevis and the lumbricals/quadratus plantae. Clinically, these orthoses have been used with children with CP and improved functional skills, such as gait and gross motor skills, have been observed. This is likely due to less restriction around ankle and possibly increased feedback through the raised sections. It was hypothesised that families would prefer the SMotOs over the AFOs for over 50% of the feedback. The objectives of this qualitative research were to:

1) Identify common themes within QoL and function relating to lower limb orthoses in children with CP.
2) Identify the impact of SMotOs and AFOs on function and on QoL in children with CP.
3) Evaluate the impact of SMotOs and AFOs on function and on QoL in children with CP.
4) Determine possible future research areas into SMotOs and AFOs and their role on function and QoL in children with CP.

7.2. Methodology

Ethical approval was obtained by Bond University Human Research Ethics Committee (RO-1835).

7.2.1. Design

The Standards for Reporting Qualitative Research was followed (O'Brien, Harris, Beckman, Reed, & Cook, 2014) when writing this chapter. A qualitative phenomenological approach was employed through a questionnaire-based survey. The questionnaire (Q'AIRE') was designed to establish the effect of lower limb orthoses in current day to day QoL. This Q'AIRE was also undertaken to determine how the SMotO and AFO affect the child's function, as reported by parents. The basic areas of focus in the Q'AIRE were chosen through discussion with families and further refined after review of other paediatric questionnaires. The initial Q'AIRE was developed and distributed to four key clinicians who were active in the area. Once the clinicians provided feedback, a pilot was conducted. From there, the final Q'AIRE was created and implemented in this study.

7.2.2. Participants

Recruitment and inclusion criteria: Twenty-three parents with children with CP who were selected for recruitment through two private therapy practices (Therapies for Kids and NAPA Centre, Sydney, NSW, Australia). Families were deemed eligible for recruitment if they had a child with CP and were using (or had used) AFOs and SMotOs. Participants were eligible from all levels of motor ability, including those with or without walking aids.

7.2.3. Instrumentation

A self-administered 24-item Q'AIRE (Appendix D, Section vi.) was designed and developed specifically to focus on AFOs and SMotOs. The Q'AIRE consisted of two sections; section one relating to AFOs and section two relating to SMotOs. Section one consisted of 18 questions with multiple choice or yes / no answers. Section two consisted of six questions with multiple choice or yes / no answers. There was an option in both sections whereby parents / caregivers could provide further information or express opinions. The Q'AIRE was designed to delve deeper into the parents/ caregiver's opinions and viewpoints on both orthoses, how they found the orthoses impacted quality of life and function for their child.

Prior to administering the Q'AIRE for parents, the tool was reviewed by two senior research academics to assure quality of questions. Fink (2005) recommends this process as a means of increasing survey reliability. The purposes of the reviews were to (a) evaluate the effectiveness of the survey for capturing required data; and (b) obtain feedback regarding the investigator-designed questions. The feedback provided by reviewers were considered against the intended scope of the research questions in order to inform revisions to the Q'AIRE for parents' tool and as such would maximise the external validity of the information captured, and the results generated by the questionnaires; a method advised by Fink (2005).

7.2.4. Procedures

An email to each participant and included an explanatory statement, a consent form and the Q'AIRE. A return date of two weeks was specified in the email to aid response timeliness. Participant assurance was provided in the explanatory statement and consent form of the participant's rights for privacy, confidentiality and anonymity. A checklist was kept monitoring the sent Q'AIRE and the returned forms. Only when the final date specified in the email had passed, was a reminder email sent, allowing one more week to complete the Q'AIRE and forms. Once the final deadline had passed, there were no further reminder emails sent, and all those who had not returned the forms were excluded.

7.2.5. Data Analysis

The qualitative data from the Q'AIRE was collected by the primary researcher through Q'AIREs returned via email. The principal investigator manually reviewed and inputted the qualitative data into an excel spreadsheet, allowing them to derive common themes and patterns, whilst a second investigator a) undertook data cleaning (accuracy verification) and b) reviewed the content analysis to ensure consistency (Boyatzis, 1998). The principal researcher read through the responses and narrative feedback families provided and identified several phenomenological themes. These themes were derived through collating the similar responses and determining the prominent themes. Two senior researchers reviewed these themes. Along with the focus of the specific Q'AIRE questions, direct quotes were used to identify recurrent or significant themes. Comparisons were made between the identified qualitative data themes were then further modified and compressed to create the final themes; aiming to reduce reviewer bias and provide a reliable and valid thematic content analysis (Gough, Yohannes, Thomas, & Sixsmith, 2013).

7.3. Results

There were 16 families, of which included local and interstate families, who consented and returned the Q'AIRE (see Figure 13). The response rate was 69.57% (16/23).

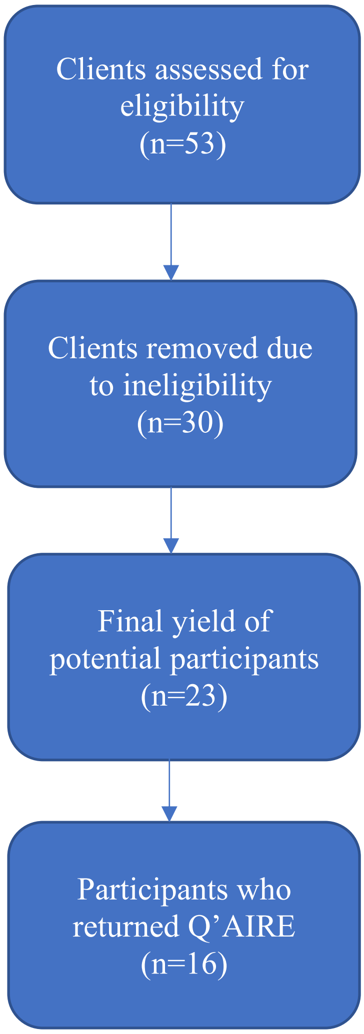


Figure 13: Flow chart of recruitment of participants for the Q'AIRE

7.3.1. Demographics

7.3.1.1. Participants

The mean age of the children when they started wearing AFOs was 2.5 years (range 1.5-5 years). The participants type of CP consisted of: spastic quadriplegia (n=5), spastic diplegia (n=5), hypotonic quadriplegia (n=2), dystonic quadriplegia (n=1), two spastic dystonic quadriplegia (n=2) and hemiplegia (n=1). The Gross Motor Function Classification System (GMFCS) Levels of participants were distributed as: GMFCS II (n=4), GMFCS III (n=6), GMFCS IV (n=5), and GMFCS V (n=1).

7.3.1.2. Orthoses

The participants of the Q'AIRE reported 81.3% (13/16) were prescribed SAFOs, 18.8% (3/16) were prescribed HAFOs and one child was prescribed a DAFO (supramalleolar). There was one family prescribed both a SAFO and HAFO and one family only used a new type of AFO (from the USA). All parents report that there were no videos taken or pedograph measurements taken pre-post AFO prescription. Follow up appointments for fittings or adjustments were had by 75% of the families. Pre-prescription appointments of SMotOs include gait observation through videos as well as foam box casting and pedographs. During SMotO fitting, the gait was observed again through video observation and any subsequent adjustments are made. Appendix E: Results of Q'AIRE provides the amalgamated responses regarding AFOs and SMotO respectively.

7.3.2. Themes

There were seven identified qualitative data themes that were compressed into four main themes. These four key areas that focussed on main components of parental feedback and consisted of: 1) time, 2) reason, 3) function and 4) comfort and dislike.

7.3.3. Theme 1: Time

The theme of 'time' reflected the wear of, and ratio between, the two orthoses. Half of the participants (50%) report wearing both orthoses. The time split between the two orthoses were (AFO/SMotO); two 30/70, two 70/30, one each of 60/40, 80/20, 20/80, 5/95. Of the remaining participants, 43.8% (7/16) did not wear AFOs anymore and one only wears AFO.

Five participants (31.3%) had been wearing the AFOs for over three years, with three other participants (18.8%) wearing the AFOs between one and three years. Only one family (6.3%) reporting to use AFO only. The type of AFO they were prescribed was of high quality from the USA and was fitted post selective percutaneous muscle lengthening surgery, therefore essential to recovery. Six families (37.5%) report their child has ceased wearing their AFOs.

Seven families (43.8%) report use of SMotOs for less than one year. Eight families (50%) report their child has used the SMotOs for between one and three years, and only one (6.3%) participant reporting use of over three years. There was 87.5% (14/16) of families reporting they preferred to use the SMotO.

7.3.4. Theme 2: Reason

'Reason' for prescription of AFOs was uncovered as an important theme in the Q'AIRE. All parents report a reason was given for AFO prescription. Alignment (43.8% or 7/16) was the most common reason for AFO prescription. Tightness in calves/ankles (stretch) and walking ability was the next most common reason for ankle prescription (37.5% each). The most interesting reason six families (37.5%) were given when prescribed AFOs was 'because the child has CP.' Five (31.3%) were told balance will be helped by AFOs. Two (12.5%) were told it will help reduce ankle rolling and reduce pointing of toes.

Parents noted there were no alternate orthosis options offered in the public health system and did not have alternate options until the SMotOs were offered in a private practice setting. It was 100% agreed across all families that they had wished there were alternate options given to their child, especially with regard to educating parents, pricing and options.

> *"I feel that there should be more options available in the public health system as the SMotOs are very expensive and need replacing every 6 months or so. Parents of kids with special needs are constantly being asked to spend money or equipment and it can become overwhelming and actually force parents to make a choice which may not be best for their child, i.e., no more SMotOs as a possible outcome! I guess the NDIS may help with this in the future. I also feel there are a lot of conflicting opinions about which is best for the child and it can be very hard for parent to make an informed decision as everyone seems to offer differing opinions."*

This opinion was similar to another parent.

> *"For children with CP - it appears there is a standard practice / framework for which children are expected to have / need. AFOs are one of these. I had to suggest my child transition from solid AFO to hinged AFO. It wasn't suggested to us. They provide better support and ankle flexibility."*

One mother reported another poor experience from the public system, stating;

> *"(the) hospital system (was) very rigid in offerings. Quite forceful if you disagree. Not progressive. AFOs had no arch support."*

7.3.5. Theme 3: Function

Improving function was a strong theme in both the AFO and SMotO feedback. The most common functional improvements when wearing AFOs families noted were improvements in balance when standing still (50%), alignment in their legs (50%), and two families (12.5%) reported improvements in transitions into standing. One family reported participant was straighter when strapped into a standing frame or wheelchair in AFOs and another reported AFOs stopped the feet rolling slightly. Improvements in walking were seen in 7 children (43.8%). These improvements were broken down further into distance (42.9%), endurance (18.2%), balance (42.9%) and speed (18.2%).

Families were asked to report on how they thought AFOs benefited their child with regard to the following: alignment (50%), balance (43.8%), calf length/stretch (37.5%), walking ability (31.3%), prevented surgery (6.3%). One family reported they felt benefit through reducing the ankle rolling slightly which gave a slightly better step and one reported benefits through support to the carer with transfers. One family (6.3%) reported ceasing the use of AFOs as the participants walking became worse compared to walking wearing SMotOs.

When wearing SMotOs, the parents of the participants noted improvements in alignment in legs (50%), balance when standing still (43.8%), sitting on the floor (43.8%), transitions into standing (37.5%) and crawling (12.5%). Improvements in walking was seen in 68.8% (11) of participants, which was further broken down into speed (81.8%), distance (72.7%), balance (54.5%) and endurance (45.5%). Five families (31.3%) reported other improvements such as improvement in fluidity of gait, development of the foot arches and improvement in foot structure. Another family reported the participant rolls less in the SMotOs and seemed to have slightly better foot placement. One family also reported that their child had better sensation and feedback as her feet were more aligned with transitions and she demonstrated improved foot placement with stepping. They also noted it was easier to change her whilst wearing SMotOs as they need to remove all clothes, which was hard with AFOs on.

Nine families (56.3%) did not notice a difference in inclusion when wearing the SMotOs vs the AFOs. Of the 43.8% that did notice a difference in inclusion when wearing SMotO, 57.1% reported this was during play with friends / siblings and inclusion in activities in school. 28.6% reported therapy was different in SMotOs than

AFO. Other differences noted in SMotOs was they were easy to fit into different shoes for different occasion, discreet and comfortable and one reported their child interacted more with others when wearing SMotOs.

7.3.6. Theme 4: Comfort & Dislike

Overall, the feedback about comfort and dislike specific to each orthoses was varied. When asked if their child was comfortable wearing their AFOs, 43.8% reported a mix of comfortable and uncomfortable, 31.3% reported comfortable, 12.5% report their child is uncomfortable and 18.8% report their child refuses to wear them. Pain was experienced by 18.8%, and 50% reported sometimes their child experiences pain. The locations for pain / discomfort as reported by parents were: feet, toes, ankle/arch and top/back of calf where they rub against the skin. Parents reported pain when the AFOs were getting too tight or small as well as not being sure where the pain is (child cries when wearing AFOs). Discomfort affected or dictated if the child wore the AFOs in 50% of the families (12.5% reported N/A).

One family reported;

> *'(Child's) high tone and dystonia meant they could push out of them (AFOs), their heel would move, and then the hard plastic pushed everywhere it shouldn't.'*

Fifteen families (93.8%) disliked the restriction in movement from the AFOs. Ten families (62.5%) disliked the way the AFOs fit into shoes, nine families (56.3%) disliked the bulkiness of the AFOs, and four families (25%) disliked the way they look. Eight families (50%) reported pressure areas from wearing AFOs and six families (37.5%) reported difficulty putting AFOs on. Three families (18.8%) reported exclusion from activities or participation at school when wearing AFOs. Families were given the opportunity to expand on their answers and reported they disliked the lack of sensory input, and lack of calf and ankle muscle development when wearing AFOs. Two families reported disliking the negative impact on function when their child was wearing AFOs, such as inability to cross legs when sitting on the floor and inability to take steps.

With regard to SMotOs, five families (31.3%) disliked the way they fit into shoes, with the responses relating to the 'bump' under the toe and the difficulty parents found with ensuring the toes were not curled under and the heel was in place. Two families (12.5%) reported they disliked the discomfort of the SMotOs with seven families (43.8%) reported their child experienced pressure areas. One family expanded this answer by describing there is some wear time to 'break in' the orthoses to become comfortable.

Families were given opportunity to expand on their answers and four families reported they felt as though the orthoses alone did not give enough structure or support around the ankles, as well as not encouraging heel down.

One mother reported that her son "*has an out-turned right foot which I have been advised is better aligned using AFO than with Piedro (shoes) and SMotO and have been advised that if we don't used AFO that his foot deformities will be worse.*"

Five families did not report any dislikes with regard to their child and SMotOs.

7.3.7. General Feedback on Orthoses Use

In those families who returned the Q'AIRE, some children use both orthoses, depending on situation, and some families who only use one type of orthoses (Table 22).

Table 22: Parental feedback surrounding SMotO and AFO

<table>
<tr><th colspan="2">Feedback from Q'AIRE</th></tr>
<tr><th>SMotO</th><th>AFO</th></tr>
<tr><td>"The SMotOs have been great for the stepping, sit to stand, pull to stand. Anything where he gets to feel the ground with the ankle movement has been the biggest bonus. Once I get some more supportive shoes to go with these then this will be the best. His Piedros still weren't helpful but we are looking at custom made ones to help this."</td><td>"We found the AFOs were pretty generic, and our daughter could easily pull out of them. We were constantly trying to add Velcro, padding etc and it was very distressing for her in the casting process, as well as putting them on each day. She would literally scream the house down."</td></tr>
<tr><td>"The SMotOs have enabled my daughter to stand with heels closer to the ground and to walk up and down stairs more easily."</td><td rowspan="2">"With an AFO, my child would drag his foot, and not walk, heel to toe. He only wears one AFO as he has right hemiplegia."</td></tr>
<tr><td>"Son is much more comfortable in SMotOs and finds it easier to manoeuvre his body and is much more willing to get up and try new things with them on because they're not as bulky."</td></tr>
<tr><td colspan="2">"I think that both have a role to play. We initially had good results with the SMotOs, but son's hips have become worse and adductors tighter than ever. His right leg has shortened and the SMotOs couldn't compensate for this, so we have had to increase time in AFOs recently. We will definitely continue to wear both going forward and change between them depending on activities. Son will be having adductor release surgery in the coming months and I expect that this will help with this hip alignment and reduced scissoring, so we will be able to use the SMotOs more."</td></tr>
<tr><td colspan="2">"We have recently gone back to wearing AFOs full time. I believe the SMotOs were great for building strength and encourage a truer walking gait. But we believe that they didn't serve the purpose of slowing down contractures which is what AFOs offer. I feel this is more important for a young child with spastic diplegic CP, the AFOs offered in Australia also have their downfalls. We feel they are very outdated and serve more to brace the child as opposed to encourage correct gait and encourage good foot posture and movement."</td></tr>
</table>

7.4. Discussion

The hypothesis that families would prefer the SMotOs over the AFOs for over 50% of the feedback was confirmed. Comparing AFO and SMotO, through the four themes, identified many positive and negative aspects for both types of orthoses from parents and caregivers of children with CP who wear AFOs and/or SMotOs. Families are searching for viable reasons and education from professionals surrounding orthosis prescription, whereby they can be empowered to assist with decision making for their child's health. It appears that as there is only one type of orthosis prescribed in the public system, and the feedback from this Q'AIRE suggest that families are looking elsewhere for other options. The most interesting reason given when prescribed AFOs was 'because the child has CP,' which may imply blanket prescription or reduced clinical reasoning.

One valid reason AFOs are prescribed for is to enhance function (Figueiredo et al., 2008). Parents report improved function and benefit with AFO across several areas. Comparatively, when asked about the SMotOs, it appeared they present a larger positive impact on function and inclusion. As function is an integral part of QoL, lower limb orthoses prescription would need to ensure enhanced function as well as alignment and comfort.

As per parent-reported questionnaire, Arnaud et al. (2008) reported that pain is associated with poor QoL in children with CP. This is supported by Houlihan, O'Donnell & Conaway (2004) who noted the presence and impact of pain in infants and children with significant neurological impairments have an impact of child's QoL and participation. In another study (Varni et al., 2005), it was found that children with CP had a lower health related QoL than healthy children and suggest that health providers should obtain health related QoL perceptions whenever possible. Relating to the presence of comfort / dislike, it appeared that SMotOs tend to have less discomfort overall than AFOs, which may be a factor in the higher compliance use of SMotO. Seven families reported pressure areas from SMotOs due to the height of the 'bumps.' Dickinson et al. (2007) concluded that physical impairments (3%) and presence of pain (7%) were responsible for the variation in QoL, and due to these findings, a child's pain should be carefully assessed. To support a good QoL and participation, both these orthoses would need to remove likely discomfort to eradicate poor compliance with wearing orthoses.

Some families reported slightly improved ease of use with SMotOs vs AFOs, which may potentially improve the QoL of the parents due to the reduction of stress and concern of AFOs being fitted correctly. Arnaud et al. (2008) noted that parents with higher levels of stress were more likely to report poorer QoL for their child across all domains. Ease of use may also tie into timeliness of getting the child ready or dressed throughout the day, which busy parents and carers may appreciate.

Parent's feedback may not necessarily hold much relevance in research studies as it is based on opinion, which may have bias. Parents tend to be the gatekeeper to intervention through therapy or equipment for their child. The responses in the Q'AIRE suggest they like to be informed in the health care decisions of their child. It appeared that all parents participating in the Q'AIRE wished the public system provided alternative options and education about orthoses. Many parents did not feel as though there was a valid enough reason for AFO prescription, so improving on clinical reasoning communication to families would appear to be important for future therapists. Providing valid options for families may also give a sense of autonomy and control with regard to intervention.

7.4.1. Limitations

This study utilized a small sample group of 16 families who were using / had used both types of orthoses. Families had children with varying severity and functional levels of CP, but there was not a strong equal dispersion in the recruited families. Despite the low numbers in each level of CP, the overall feedback was similar across the board.

7.4.2. Implications

The implications of this Q'AIRE encourage further research to determine impacts of orthoses on QoL in children with CP. A more in-depth, longitudinal study including an extensive QoL questionnaire on a more homogenous sample could be beneficial to determining the effect of orthoses in a qualitative sense. Future studies could include randomized controlled trials examining AFOs and SMotOs, using significant sample size, a larger range of GMFCS levels and more varied types of CP to determine how the population responds to each orthoses (if there is a 'better' prescription option depending on type and severity of CP) and developing a wearing timeline between two orthoses. Allowing wear time between each orthosis allows for the benefits of both, e.g., a stretch and stability from AFO, whilst the SMotO allows for dynamic ankle movement and sensory feedback. The results of this Q'AIRE may also serve to bring awareness to clinicians of the effect orthoses may have on QoL of children with CP and their families.

7.5. Conclusion

From qualitative parental feedback, it appeared there was more preference to wear SMotOs for comfort, function, and in general, as preference to traditional AFOs.

This qualitative Q'AIRE study creates a base for further investigation and research into the impact on QoL of orthoses used in children with CP through a larger cohort, as well as studies on long-term impact of both AFOs and SMotOs. It may also be of benefit to implement educational training for prescribers with regard to clinical reasoning communication, and how to provide orthotic options more effectively to parents.

8.0. Sensomotoric Orthoses, Ankle-Foot Orthoses, and Children with Cerebral Palsy: The Bigger Picture

Prelude

The specialised population (paediatric cerebral palsy) that informed this program of research was complex to assess and analyse, due to the inability of the children to follow directions of some assessment tools (comprehension, physical or behavioural difficulties). Therefore, a smaller than expected amount of quantitative data were collected. Despite this, a large amount of qualitative information was gleaned from the parents in the questionnaire as to what was deemed most important to their child and their daily functioning. This case series aims to encapsulate the last four studies by putting together the qualitative and quantitative data, imagery and videography to provide a complete picture of the child with cerebral palsy and the use of the nominated orthotic devices. This case series was published in the open access journal Children*. Please find the full PDF version in Appendix B.*

8.1. Introduction

Ankle-foot orthoses (AFOs) and sensomotoric orthoses (SMotOs) have been described in depth throughout this thesis. The benefits of using AFOs in children with cerebral palsy (CP) has been well documented over the years. The SMotO is a clinically relevant, yet under researched, orthoses option used in the same population.

Previous chapters (Chapters 3, 5, 6 and 7) have investigated the effect of these two orthoses on gait and gross motor skills in children with CP as well as the qualitative effect on life for them and their caregivers through a questionnaire (Q'AIRE). The systematic review (Chapter 3) critically appraised studies surrounding lower limb orthoses and the effect on gait and gross motor skills. Lower limb orthoses (AFOs and postural control insoles), particularly the more dynamic orthoses, were found to improve gait and gross motor skills in children with CP. Studies 5 and 6 (reported in Chapters 5 and 6) investigated the effects of SMotO and AFO on gross motor skills and gait in children with CP. The findings reported in these two studies were that the SMotO improved gait and gross motor skills, but further studies will need to be performed in order to support these results with larger participant numbers. Through the qualitative Q'AIRE, Study 7 (Chapter 7) investigated the effect on quality of life when children with CP wore AFOs and SMotOs from the parents' / caregivers' perspective. This study found there were more positive feedback points relating to SMotOs versus AFOs throughout the four key theme areas: Time, Reason, Function, and Comfort and Dislike. Other than the current thesis chapters, there is no other research into the effect of SMotO on gait, gross motor skills and quality of life in children with CP. To provide a more complete picture of these complex children, a need to merge these studies of a select group of participants was found.

Creating a 'real life' picture of particular 'cases' or participants in a mixed method case series study can bring depth to understanding both the clinical relevance and impact of an intervention on certain aspects of life. Although case series represent a low level of evidence (IV) and have methodological limitations with regard to making causal inferences about the relation between treatment and outcome (Kooistra, Dijkman, Einhorn, & Bhandari, 2009), Murad et al. (2018) suggested that when no other higher level of evidence is available, decision-making can be informed using evidence derived from case reports and case series.

Therefore, noting the lack of literature in this field (See Chapter 3), the aim of this case study series was to synthesis and enrich the volume of evidence reported in this thesis to inform real world application of SMotO use in children with CP. Presenting the case series demonstrated the impact of SMotOs and AFOs on function, movement and quality of life in the individual, in a way that is clinically relatable.

8.2. Methods

8.2.1. Participants

Recruitment and inclusion criteria: Participants were children with CP recruited by convenience sampling through two private therapy practices (Therapies for Kids and NAPA Centre, Sydney, NSW, Australia). The inclusion criteria were: a) diagnosis of CP with any Gross Motor Function Classification System (GMFCS), b) using SMotOs / AFOs (or have used them) and completed the wearing in process, and c) no surgery in past six weeks.

8.2.2. Study Design

Ethical process: Ethical approval was obtained through the Bond University Research and Ethics Committee (Approval RO-1835). Consent was gained from Clinic Directors in both private practice settings. Parents / caregivers were given an explanatory statement and consent form, both of which were read and completed before data collection took place.

This study was a retrospective mixed method design, with a combination of both quantitative and qualitative outcome measures collected. Outcome measures were undertaken in two separate clinic locations, as well as six home settings due to the families being unable to travel. These settings were selected as they were familiar to the child and allowed the parents and/or siblings to be present throughout the testing. Relevant, pertinent participant qualitative information from the Q'AIRE was extracted and combined with the correlating participants quantitative measurements to create the case series and is described in greater detail below.

8.2.3. Intervention

All participants brought their own SMotOs and AFOs. The AFOs were custom-made through public or private orthotists from polypropylene with Velcro straps holding the foot in place. The AFOs all had a full-length foot plate. Due to the AFOs being the participants 'usual prescription' there was no further assessment or measurements of the AFOs performed. The SMotOs were custom made for each child from ethyl vinyl acetate (EVA) and had been assessed and prescribed by a podiatrist or pedorthist who were both expert in this design type of orthosis. Each participant used SMotOs and/or AFOs whilst participating in outcome measures.

8.2.4. Quantitative Outcome Measures

The quantitative section of the case series process included the principal researcher assessing each participant as able in the: Timed Up and Go (TUG), Berg Balance Scale (BBS), The Gross Motor Function Measure (GMFM), and / or the Edinburgh Visual Gait Score (EVGS). Each outcome measure was performed as the child was able, in any order it seemed appropriate, and in any order of orthoses. For example, one child came in wearing AFOs and wanted to walk around the clinic, therefore the EVGS in AFOs was assessed first. This child was then becoming interested in some static activities, therefore the GMFM in AFOs was performed next. Data collection continued for as many outcome measures as was possible for each of the participants' and within their ability and tolerance.

8.2.5. Qualitative Outcome Measures

Three styles of qualitative evidence were included: written feedback from parents compiled from the Q'AIRE, and photo imagery of pedographs (pre-and post SMotO) and / or video imagery of gait.

The Q'AIRE was emailed to multiple families as per Study 7 (reported in Chapter 7) after participating in the data collection. There were eight participants included in this case series. The themes were identified as previously described in Chapter 7. Qualitative video imagery of a typical gait pattern was taken with the participant barefoot (where able), in AFOs and SMotOs. Video imagery was taken with a handheld device (Apple iPhone 7s, Apple Inc., CA, USA). The videos were taken in whichever location the outcome measures were recorded – either in clinic or at the participants home - whilst the participant mobilised at a self-directed pace, using their usual prescribed walking aid (where necessary). The podiatrist and pedorthist who prescribed and fabricated the SMotOs provided pedograph images of two participants (7 and 8) footprints before and after the use of SMotOs.

8.3. Results

8.3.1. Participants and Outcome Measures

Data for eight participants (male: n=7: female: n=1) were collected. Participant 2 had EVA heel wedges on their solid AFO to encourage weight-through-heel, mimicking heel strike. The eight participants demonstrated a large range of physical ability with reported GMFCS levels of I (n=1 participant), II (n=2 participants), III (n=2 participants) and IV (n=3 participants). The age range was three to 13 years (average age = 7 ± 3.7 years). Overall, there were 39 quantitative and six qualitative measures collected (Table 23). The EVGS demonstrated the highest response. Please note in videos participants were previously coded (embedded in video) and as such, may display a different participant number to current number.

Table 23: Participant quantitative and qualitative outcome measures response

Outcome Measure		**Intervention (n=)**		
Quantitative		SMotO	AFO	Barefoot
	EVGS	7	6	2
	GMFM-88	5	5	0
	BBS	4	4	0
	TUG	3	3	0
Qualitative		**Responses (n=)**		
	Q'AIRE	4		
	Pedograph	2		
	Videography of gait	6		

8.3.2. Case Series

The presentation of each of the eight retained participant data is presented below as individual cases.

Participant 1: 4-year-old male child with spastic diplegic CP, GMFCS **III.** Mobilises with reverse walker.

Outcome Measures:

Participant 1 demonstrated better scores in TUG, GMFM-88 and EVGS when in SMotO, likely due to the dynamic nature of the SMotOs being used in dynamic outcome measures (Table 26). Similar to the findings within Chapter 5, Participant 1 displayed a better score in the BBS when in AFOs, likely due to the bracing effect of AFOs.

As per response from Q'AIRE, the participant's Mother reported, *"I have been advised by some of our health care professionals that (my) son's gait is better in his AFOs than in Piedro (supportive disability shoe) with SMotO."* This statement is contradicted by the EVGS results (Table 24). Participant 1 did not give consent for video imagery of gait.

Table 24: Participant 1 outcome measure comparative results between SMotO and AFO

Outcome Measure	SMotO	AFO
TUG (sec)	13.8 sec	17 sec
BBS (/56)	15	12
GMFM-88 (%)	73.51	71.17
EVGS	25 (total L & R)	38 (total L & R)

Participant 2: 8-year-old male child with spastic quadriplegic CP, GMFCS III. Mobilises in a reverse walker.

Outcome Measures:

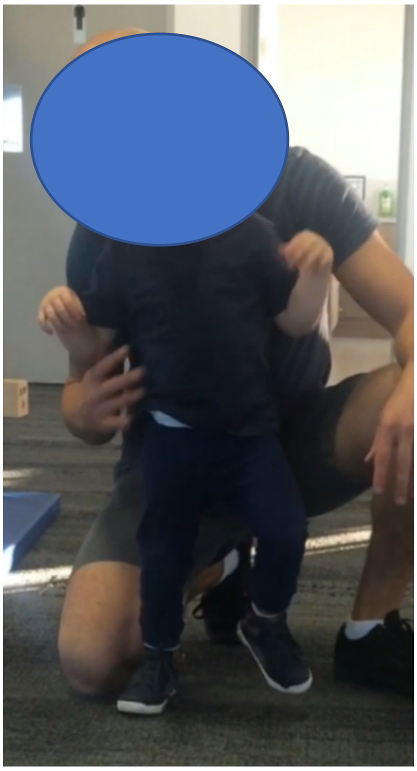
Participant 2 performed better in the TUG, GMFM-88 and EVGS when wearing SMotOs likely due to the dynamic nature of the SMotOs being used in dynamic outcome measures (Table 25). Interestingly, the BBS reported the same score for both orthoses. Correlating videos (in DropBox folder link below) highlighting gait in SMotO, AFO and barefoot (as labelled) for 'Participant 2' have been provided for reference.

https://www.dropbox.com/sh/tfcrp9c1dxwmbn0/AAB9FSgGPunYpi8uCDDxZKpAa?dl=0

Mother reported, as per Q'AIRE, *"the SMotOs have been great for the stepping, sit to stand, pull to stand. Anything where he gets to feel the ground with the ankle movement has been the biggest bonus. Once I get some more supportive shoes to go with these then this will be the best. His Piedros still weren't helpful but we are looking at custom made ones to help this."*

Table 25: Participant 2 outcome measure comparative results between SMotO and AFO

Outcome Measure	SMotO	AFO
TUG (sec)	41.13	44.37
BBS (/56)	7	7
GMFM-88 (%)	69.11	64.41
EVGS	30 (total L & R)	41 (total L & R)



Participant 3: 4-year-old boy with spastic diplegic CP, GMFCS II.

Outcome Measures:

Participant 3 demonstrated improved scores in the BBS, GMFM-88 and EVGS when wearing SMotOs compared to AFOs (Table 26). The GMFM-88 demonstrates a change of 6% which is reported as a clinically important change in score.

Correlating videos (in DropBox folder (link below) highlighting gait in SMotO, AFO and barefoot (as labelled) for 'Participant 3' have been provided for reference.
https://www.dropbox.com/sh/ew4mbh9elsgpk63/AACXVALqehvueHW0bVabgQHaa?dl=0

The mother of Participant 3 reported, as per Q'AIRE, that her *"son is much more comfortable in SMotOs and finds it easier to manoeuvre his body and is much more willing to get up and try new things with them on because they're not as bulky."*

Table 26: Participant 3 outcome measure comparative results between SMotO and AFO

Outcome Measure	SMotO	AFO
TUG (sec)	Unable to follow direction	
BBS (/56)	17	13
GMFM-88 (%)	85.51	79.51
EVGS	8 (total L & R)	15 (total L & R)

Participant 4: 13-year-old girl with spastic quadriplegic CP, GMFCS II.

Outcome Measures:

Participant 4 visually appeared to walk well in AFOs, but the results of the EVGS (Table 27) demonstrated a notable difference in quality of gait pattern when wearing AFOs compared to SMotO. Her GMFM-88 total score did not display a large difference in scores between orthoses, indicating that neither orthosis demonstrates an increased effect on gross motor skills compared to the other.

Correlating videos (in DropBox folder (link below) highlighting gait in SMotO, AFO and barefoot (as labelled) for 'Participant 4' have been provided for reference. Participant 4 did not complete the Q'AIRE.

https://www.dropbox.com/sh/1gd73f6b3uufwzh/AAAQM-uMtXsmpPieC86bStp9a?dl=0

Table 27: Participant 4 outcome measure comparative results between SMotO and AFO

Outcome Measure	SMotO	AFO
TUG	11.33	10.13
BBS	39.00	37.00
GMFM-88 (%)	91.29	92.00
EVGS	15 (total L & R)	31 (total L & R)

Participant 5: 4-year-old boy with dystonic quadriplegic CP, GMFCS IV. Mobilises in a supportive reverse walker.

Outcome Measures:

Participant 5 was physically affected by his dystonia and used a gait trainer to mobilise. He was unable to participate in any other outcome measures.

Despite this limitation, the video imagery and EVGS both demonstrated the clear differences in his gait between barefoot, AFO and SMotOs. The qualitiative evidence highlighting gait in the three conditions (correlating videos in DropBox folder link below) is supported by the results from the EVGS (Table 28).

https://www.dropbox.com/sh/8uolo7lhk5v5g2e/AAAV_tK_KujMt0Sew6t__p5ba?dl=0

Table 28: Participant 5 comparative EVGS results between barefoot, AFO and SMotO

Outcome Measure	Barefoot	AFO	SMotO
EVGS	51 (total L & R)	30 (total L & R)	17 (total L & R)

Participant 6: 5-year-old boy with spastic quadriplegic CP, GMFCS IV. Mobilises in a reverse walker and hip abduction brace.

Participant 6 struggled to walk without the support of his orthoses, walking frame and abduction brace.

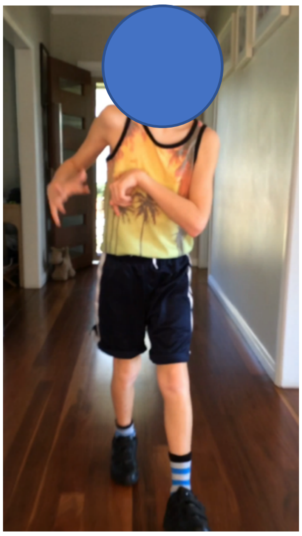
Outcome Measures:
The results from the GMFM-88 (Table 29) showed a mild difference in scores between orthoses.

Both the quantitative measure (EVGS) and qualitative imagery (video as per link below), demonstrated the difference in quality of movement between orthoses and barefoot. Correlating videos (in DropBox folder link below) highlighting gait in SMotO, AFO and barefoot (as labelled) for 'Participant 6' have been provided for reference. Participant 6 did not complete the Q'AIRE.

https://www.dropbox.com/sh/khmll6jwoumlpnv/AAAAgXhzhgX4PJGFpjRX6FR6a?dl=0

Table 29: Participant 6 outcome measure comparative results between SMotO and AFO

Outcome Measure	SMotO	AFO
GMFM-88 (%)	49.52	47.67
EVGS	9 (total L & R)	25 (total L & R)



Participant 7: 12-year-old boy with spastic dystonic quadriplegic CP, GMFCS I.

Outcome Measures:

Participant 7 was independently mobile with and without shoes. Participant 7 demonstrated improved alignment and stability when he wore SMotO as per EVGS score (Table 30) compared to barefoot. Participant 7 was not included in Chapter 6 as he no longer wore AFOs and therefore did not meet the inclusion criteria. The images from the Pedographs (Figure 14 and Figure 15) demonstrated visual weightbearing changes pre-SMotO and one year after using SMotO, especially through right foot. Correlating videos (in DropBox folder (link below) highlighting gait in SMotO and barefoot (as labelled) for 'Participant 7' have been provided for reference. Participant 7 did not complete any other outcome measures.

https://www.dropbox.com/sh/i25vyb9jnd5v6f2/AABj5pBSJ4MNBj9kk4LPDbRSa?dl=0

Table 30: Participant 7 EVGS results SMotO and barefoot

Outcome Measure	SMotO	Barefoot
EVGS	3 (total L & R)	13 (total L & R)

Figure 14: Left and right initial pedograph footprint (2016)

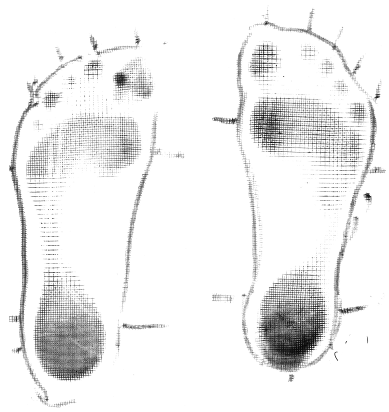


Figure 15: Left and right final pedograph footprint (2017)

Participant 8: 6-year-old boy with dystonic quadruplegic CP, GMFCS IV. Mobilises with assitance in a supported walker.

Participant 8 was originally prescribed solid AFOs then hinged AFOs despite not having any restriction in ankle range of motion. He was able to weight bear with support and walks in a walker. He uses SMotOs in a Piedro shoe.

Outcome Measures:

Participant 8 was unable to complete any of the quantitative outcome measures due to his severe dystonia. From the Q'AIRE, mother reported *"for children with CP - it appears there is a standard practice / framework for which children are expected to have / need. AFOs are one of these. I had to suggest my child transition from solid AFO to hinged AFO. It wasn't suggested to us. They provide better support and ankle flexibility."*

Figure 16 and 17 demonstrate the changes seen (over a seven month period) in the muscle activation of his foot when wearing the SMotOs. These pedograph images corroborate the theory of the muscles learning to activate and support the foot, despite his CP diagnosis.

Figure 16: Left and right initial pedograph footprint (15/06/2016)

Figure 17: Left and right final pedograph footprint (24/01/2017)

8.4. Discussion

Collecting a range of data in this population is challenging due to age, level of disability, cognitive comprehension, inability to process instructions or give feedback, poor motor planning, and general behaviour. As such, data can often be incomplete when multiple outcome measures are collected. Given the challenges of collection and the volume and variety of data collected in this program of research, the use of a mixed method approach allowed for the collation of both quantitative and qualitive data to enrich the research findings. Furthermore, the inclusion of qualitative information to create a more holistic viewpoint to the intervention findings allowed the caregivers to validate and express their experiences.

The individual outcome measure results suggested a difference between areas of static and dynamic movement with SMotO and AFO. The general observed trend was increased static balance whilst wearing the AFO, and general improved ability (score) with dynamic movement when wearing the SMotO. This result is encouraging as a foundation to warrant further investigation into the use of SMotOs in this population for gross motor skills.

It appears, through both the qualitative and quantitative results, that children with CP have some preference to using the SMotOs. In support of this, when looking at the Q'AIRE qualitative data (Study 7 in Chapter 7), parents tended to more positive comments regarding the use of SMotOs when compared to AFOs for gross motor skills and ease of use. In addition, it was identified that families do not have follow up appointments to reassess gait with AFOs and the impact of the AFO on gross motor skills. Clinically, it may also be of benefit to implement a follow up timeline to reassess the effect of orthoses prescribed.

Using a clinic where a large number of clients reside interstate or internationally, leaves the study somewhat lacking a significant number of potential participants, hence the low numbers of participants and inconsistent numbers of completed outcome measures. Some families had agreed to participate in all areas of the study but were unable to commit to the time required to complete the assessment, unable to participate due to distance or were unable to complete assessment due to the child's behaviour. A limiting factor of participants completing all outcome measures was the inability of the child to comprehend complex instructions relating to outcome measures or participation was affected due to lack of processing verbal instruction.

8.4.1. Future Research Direction

Acknowledging that this small case series demonstrates a minute part of the population, it does provide some important insights into the child as a ‘whole picture’ versus statistically based evidence alone. Future research could be to create a more specific assessment to determine the ability of the child, the family goals, and to investigate the in relation to the ability of SMotOs and AFOs to meet these goals. The idea being a more individualised, child-centred, goal-driven approach to orthoses prescription.

8.5. Conclusion

The aim of the current case series was to synthesise different participants’ qualitative and quantitative evidence to support the volume of evidence in this thesis. It was also aimed at informing real world application of SMotO in children with CP using clinically relevant outcome measures relating to gait and gross motor skills orthoses prescription and what may be the best course of action for families and health professionals.

It was concluded, based on the overall results of this case series, that a general improvement was seen in gross motor skills and gait when wearing the SMotOs compared to AFOs and the SMotOs were preferred by some parents. This case series added depth to the thesis by tying all the information gathered in previous chapters into individual presentations. By displaying video imagery of participants’ gait in AFO, SMotO and barefoot, the viewers were able to correlate quantitative results to qualitative performance.

9.0. Thesis Discussion

Prelude

This chapter serves to provide a summary of the research undertaken through which to inform this thesis and, specifically, connects the aims and hypotheses with the key findings of each study. This chapter provides a summary of each study and some new information for therapists to consider when prescribing lower limb orthoses in children with cerebral palsy. Finally, this chapter also provides study limitations, clinical implications, and areas of future research, as well as an overall thesis conclusion.

9.1. Summary of Aims & Hypotheses

The overarching of the aims and hypotheses of this thesis were to;

a) introduce paediatric physiotherapy and the background of cerebral palsy (CP);

b) investigate the reliability of the Anterior Line Method (ALM) through a proof-of-concept experimental design study and to further explore the use of this method in the paediatric population. It was hypothesised that the ALM would be more reliable when implemented by more experienced assessors;

c) systematically review the literature orthoses, with a specific focus on children with CP, and to explore the effects lower limb orthoses have on improving gait and gross motor skills (GMS). It was hypothesised that lower limb orthoses would improve alignment, cadence, and balance;

d) explore the feasibility of several outcome measures used in a timely and effective manner to assess gait and GMS in children with CP while wearing ankle-foot orthoses (AFOs) and sensomotoric orthoses (SMotOs). It was hypothesised that three out of six outcome measures would be feasible to implement in a timely manner (twice), and that gait and GMS would be improved when wearing SMotOs more than AFOs; and

e) display and compare qualitative analyses surrounding quality of life (specific to gait and GMS) in children with CP when wearing two different lower limb orthoses, with the hypothesis that families would prefer the SMotO over the AFOs for over 50% of the feedback.

9.2. Summary of Key Findings

The first study to inform this thesis investigated the reliability of the ALM, (an alignment assessment method for the lower limb) finding that the reliability of the ALM was greater when implemented by trained experienced assessors and practitioners. When assessors were less experienced, reliability was more adversely impacted in children than adults. However, it was also identified that this method may have value in a typical paediatric population assessment when experienced assessors were applying the method. Certain components of the ALM (resting calcaneal stance position) were more reliably assessed than others (neutral calcaneal stance position). This study, however, had several limitations and as such, the ALM would benefit from further investigation into application and reliability. Despite the lower reliability, the ALM was found to be regularly used in some paediatric clinics, especially when in combination with other techniques to assess children's lower limb alignment, specifically tibial torsion.

Clinically, typical low-profile orthotics were used for children with issues in lower limb alignment, e.g., pronated feet or in-and out-toeing. However, for children with CP, varying styles of AFOs (e.g., solid- AFO, hinged-AFO, dynamic or supramalleolar AFO) were the orthoses seen in the clinical practice. As the use of SMotOs had begun to emerge and be implemented in therapy clinics treating children with CP, the lack of published research employing these orthoses, highlighted the need for further investigation into these orthoses.

The original systematic review search strategy design failed to find papers investigating the use of SMotO in children with CP. Therefore, the review parameters were altered and a narrative systematic review was undertaken to discover current research on lower limb orthoses use in this population, and how lower limb orthoses improved the gait and GMS in children with CP. The seven resultant papers supported the use of AFOs and postural control insoles in improving gait and GMS in children with CP, especially those with a dynamic effect, such as a hinge or lower support at the ankle. Interestingly, using those parameters did not uncover orthoses that used tone reducing features, such as those used in AFO studies by Crenshaw et al. (2000), and Ibuki et al. (2010). Due to the lack of research on SMotO and their effectiveness on gait and GMS in children with CP, a gap in research focussing on the effect of SMotO on improving mobility and functional physical ability in children with CP, was identified.

A pilot feasibility study, performed to address this gap in the research found that, with regard to patient time and therapist skill, the Berg Balance Scale (BBS), Gross Motor Function Measure (GMFM) were the most feasible and timely for assessing changes in GMS, and the Edinburgh Visual Gait Score (EVGS) was viable for use in gait analysis, thus confirming the hypothesis.

To further investigate the use of SMotOs in the paediatric CP population, a series of cross-sectional cohort intervention studies were presented. The cross-sectional studies were performed wearing the SMotOs compared to the AFOs and using the outcome measures from the feasibility pilot study. Clinically, the SMotOs were found to have demonstrated improved foot alignment, balance, control with walking and improved functional gross motor skills. Study 5 (Chapter 5) showed evidence of improved GMS when wearing SMotOs compared to AFOs, with a medium positive change in the GMFM-88, and the largest score changes were seen in the walking and running segments. Study 6 (Chapter 6) further demonstrated increased gait quality through using the EVGS when using the SMotO compared to the AFOs. During these studies, as well as through clinical experience, parents were noting how much less invasive and obvious the SMotOs were for their child and how this affected their quality of life.

While, as clinicians, comfort and ease of use of equipment (as well as effectiveness of intervention), would support prescription of equipment and orthoses, based on quantitative information, qualitative information presented by parents was sought to further enrich findings. When exploring the impact of these two types of lower limb orthoses on quality of life in children with CP through questionnaire four themes were created based on parent and carer responses. These were: time, reason, function, and comfort and dislike. The main findings under these themes were that it appeared to be more preference to wear SMotOs for comfort, function, and in general, as preference to traditional AFOs.

The case series was able to encapsulate the previous four chapters by merging the qualitative and quantitative data, imagery and videography to provide a complete picture of the child with cerebral palsy and the use of the nominated orthotic devices.

9.3. Summary of Study Limitations

Due to the nature of the participants investigated throughout the thesis, there were several limitations to these studies, such as;

- Small sample sizes: Recruitment yields were typically small. This was most likely due to the inability of parents / caregivers to commit to attend, the participants inability to complete the required tasks, and the complex nature of the clients at the clinic (for example, the high level of complex diagnoses in the clinics meant there were a notable number of children excluded due to not meeting the inclusion criteria for diagnosis). Similar sample sizes in this population have been used in other studies investigating gait in CP (Lee et al. 2008). Despite the sample size, the preliminary results were promising.

- study designs; to properly compare the effect of the orthoses, including barefoot comparisons would have demonstrated a baseline score of each participant. Including the extra comparison was not feasible for this population as completing six rounds of outcome measures would have been too tiring, both mentally and physically, as well as some participants not being able to mobilise barefoot. Noting this study design limitation, the approach taken in this study did allow for the ability to capture data that was less likely to be confounded by fatigue.
- not investigating the long-term effect of the orthoses on the outcome measures. However, the study protocols and outcome measures used provide a platform for future research to build upon.
- the specialised population that informed this program of research was complex to assess and analyse, due to the inability of the children to follow directions of some assessment tools (comprehension, physical or behavioural difficulties), therefore a smaller than expected amount of quantitative and qualitative data were collected. However, as several of these outcome measures and assessment tools are validated for use in this population, the complexity of the participants provides an inclusive series of preliminary results to inform future studies.

9.4. Clinical Application and Relevance

As outcome measures are used in clinical settings for assessment and prescription, the following points were found when working with children with CP:

- ALM: Using the ALM as an assessment technique for lower limb alignment in a paediatric setting may not be clinically viable unless further research and training is completed by future researchers and assessors.
- GMFM-88:Utilising gross motor assessment techniques in a typical paediatric physiotherapy clinic setting, the GMFM-88 was found to be relatively easy to employ to assess the participant's physical ability. The GMFM-88 was particularly relevant for more affected children, as

some of the tasks were completed without verbal, receptive or expressive language. Rather, the participant was cued through motivation to reach for a toy or was placed in a position to assess movement out of the position.

- The BBS appeared to be a little more complex to employ as participants had to follow specific directions. It may be more clinically relevant to use alternate balance scale measures when assessing children with cognition difficulties.
- EVGS: Using an observation scale for gait assessment is a clinically relevant tool to use, especially when the clinic is not set up as a gait laboratory. Ensuring the video capture is recorded as per instructions, the EVGS is a relatively straightforward assessment tool to use. It allows for timely revision of gait, as a video recording enables the clinician to spend time observing the movement and angles.

When observing parents assisting the participants don and doff the orthoses throughout the studies, the AFOs appeared to require more time and effort to apply, whereas the SMotOs appeared to be donned faster. Parents confirmed the ease of use of SMotOs in the questionnaire. This may be clinically relevant to clinicians and clients when there is a time constraint.

The clinical relevance of the studies presented in this thesis could be best summarised by noting that there are different lower limbs orthoses interventions and outcome measures that may be warranted for use with some children with CP. Taking into consideration the effect of SMotO on gait, gross motor skills and QoL, the SMotOs present a clinically viable alternate orthoses option for children with CP. Therefore, there appears to be value and evidence to further support research in this area.

9.5. Future Studies

Although the studies reported in this thesis demonstrated encouraging preliminary evidence from the quantitative results to support the use of SMotO in this population, future research will further enrich and contextualise the results of these studies through a longer intervention time, increased participant study numbers, and further refined research methods. With the ever-expanding wealth of knowledge that research provides, investigating therapeutic interventions in a thorough manner will not only benefit the clinicians, but the patients or clients they work with.

Future studies should also include long term investigations into the use of SMotOs in children with CP with a focus on how they affect gait and GMS in the long term and potentially across the lifespan. This will allow those prescribing SMotOs to better understand the long-term affects and benefits of wearing SMotOs, further contributing to quality of patient care.

In addition, the feasibility of a modified SMotO / AFO combined orthoses warrants investigation given that, when a SMotO alone cannot overcome spasticity, returning to a solid, orthopaedic shoe is recommended. To further align findings, a future study investigating the modified or combined orthoses should follow similar protocols and outcome measures as stated in Study 5 (Chapter 5), Study 6 (Chapter 6) and Study 7 (Chapter 7) in order to encapsulate the whole patient.

9.6. Conclusion

The volume of research presented in this thesis provides information for physiotherapists with regard to lower limb orthoses in a paediatric population, specifically children with CP. It clearly identifies gaps in the current research surrounding SMotOs and provides some evidence into the use of these orthoses in a clinical setting. The baseline research in this thesis allows for extensive future research into these orthoses, and the potential for significant improvements in gait, gross motor skills and quality of life in children with CP. The findings from this work can be used to advance the profession by introducing these orthoses to clinicians, providing clinically applicable and clinically relevant evidence, and potentially position stands detailing best practice for these orthoses in children with CP. Clinically relevant information is also provided to guide the holistic view of peers into how lower limb orthoses can affect the daily life of children with CP. The future possibilities of researching the use of SMotOs in paediatric populations, specifically CP, are very promising.

10. References

Aiona, M., Calligeros, K., & Pierce, R. (2012). Coronal Plane Knee Moments Improve After Correcting External Tibial Torsion in Patients with Cerebral Palsy. *Clinical Orthopaedics and Related Research*, 470(5):1327 - 1333.

Akobeng, A. K. (2005). Understanding randomised controlled trials. *Archives of Disease in Childhood*, 90(8):840-844.

Alotaibi, M., Long, T., Kennedy, E., & Bavishi, S. (2014). The efficacy of GMFM-88 and GMFM-66 to detect changes in gross motor function in children with cerebral palsy (CP): a literature review. *Disability and Rehabilitation*, 36(8):617-627.

APA. (2006). *Australian Physiotherapy Association.* Retrieved from Position Statement - Provision and Access to Aids and Equipment for People With Disabilities: http://www.physiotherapy.asn.au/DocumentsFolder/Advocacy_Position_Aid_Epuipment_Disability_2010.pdf

APA. (2008). *Australian Physiotherapy Association.* Retrieved from Position Statement - Children with Mild to Moderate Physical Disabilities in Schools : http://www.physiotherapy.asn.au/DocumentsFolder/Advocacy_Position_Children_Disability_2008.pdf

APA. (2013, August). *Position Statements - Health and Wellbeing of Children and Young People.* Retrieved from Australian Physiotherapy Association: http://www.physiotherapy.asn.au/DocumentsFolder/APAWCM/Advocacy/2018_HealthWellbeing_ChildrenandYoungPeople.pdf

APA. (2017). *Australian Physiotherapy Association*. Retrieved from Physiotherapy in Australia: http://www.physiotherapy.asn.au/APAWCM/Physio_and_You/APAWCM/Physio_and_You/physio.aspx?hkey=1448ce89-2621-4ccf-8b00-ff0d45df985d

Arnaud, C., White-Koning, M., Michelsen, S. I., Parkes, J., Parkinson, K., Thyen, U., . . . Colver, A. (2008). Parent-Reported Quality of Life of Children With Cerebral Palsy in Europe. *Pediatrics*, 121(1):54-64.

Basaran, S. H., Ercin, E., Bayrak, A., Cumen, H., Bilgil, M. G., Inci, E., Avkan, M. C. (2015). The measurement of tibial torsion by magnetic reasonance imaging in children: the comparison of three different methods. *European Journal of Orthopaedic Surgery & Traumotology*, 25(8)

Bella, G. P., Rodrigues, N. B., Valenciano, P. J., Silva, L. M., & Souza, R. C. (2012). Correlation among the Visual Gait Assessment Scale, Edinburgh Visual Gait Scale and Observational Gait Scale in children with spastic diplegic cerebral palsy. *Revista Brasileira de Fisioterapia*, 16(2):1340-40.

Bentley, I. (2012). Assessment of inter- and intra-rater reliability, and agreement of two clinical methods to evaluate tibial torsion: the Gravity goniometer and Footprint method. *Queen Margaret University*, Other thesis.

Berg, K., Wood-Dauphinee, S. L., Williams, J. I., Maki, B. (1992). Measuring balance in the elderly: validation of an instrument. *Canadian Journal of Public Health, 83(Suppl 2):*S7-11.

Best, W. (2005). Interdisciplinary team helps children to get back on their feet. *Medicine & Technology*, 28-30.

Bjornson, K. F., Schmale, G. A., Adamczyk-Foster, A., & McLaughlin, J. (2006). The Effect of Dynamic Ankle Foot Orthoses on Function in Children With Cerebral Palsy. *Journal of Pediatric Orthopedics*, 26(6):773 - 776.

Bjornson, K., Zhou, C., Fatone, S., Orendurff, M., Stevenson, R., & Rashid, S. (2016). The Effect of Ankle-Foot Orthoses on Community-Based Walking in Cerebral Palsy: A Clinical Pilot Study. *Pediatric Physical Therapy*, 28(2):179–186.

Boyatzis, R. E. (1998). *Transforming qualitative information: Thematic analysis and code development.* Sage Publications Inc.

Boyce, W. F., Gowland, C., Rosenbaum, P. L., Lane, M., Plews, N., Goldsmith, C., . . . Zdrobov, S. (1991). Measuring Quality of Movement in Cerebral Palsy: A Review of Instruments. *Physical Therapy*, 71(11):813-819.

Boyd, R. N., Pliatsios, V., Starr, R., Wolfe, R., & Graham, KH. (2000). Biomechanical transformation of the gastroc–soleus muscle with botulinum toxin A in children with cerebral palsy. *Developmental Medicine & Child Neurology*, 42(1):32–41.

Brodke, D. S., Skinner, S. R., Lamoreux, L. W., Johanson, M. E., St. Helen, R., & Moran, S. A. (1989). Effects of ankle-foot orthoses on the Gait of Children. *Journal of Pediatric Orthopaedics*, 9(6):702–708.

Brukner, P., & Khan, K. (2001). *Clinical sports medicine.* Sydney: Australia: McGraw-Hill.

Buckon, C.E., Thomas, S.S., Jakobson-Huston, S., Sussman, M., & Aiona, M. (2001). Comparison of three ankle-foot orthosis configurations for children with spastic hemiplegia. *Developmental Medicine & Child Neurology*, 43(6):371-378.

Buckon, C.E., Thomas, S.S., Jakobson-Huston, S., Moor, M., Sussman, M., & Aiona, M. (2004). Comparison of three ankle–foot orthosis configurations for children with spastic diplegia. *Developmental Medicine & Child Neurology*, 46(9):590–598.

Butler-Manuel, P. A., Guy, R. L., & Heatley, F. W. (1992). Measurement of tibial torsion: a new technique applicable to ultrasound and computed tomography. *British Journal of Radiology*, 65(770):119-126.

CanChild. (2020). *Gross Motor Function Measure (GMFM)*. Retrieved from CanChild Resources: https://canchild.ca/en/resources/44-gross-motor-function-measure-gmfm

Carey, H., Martin, K., Heathcock, J., & Comb-Miller, S. (2015). Determining the minimal detectable change and the minimal clinical important difference of the

Timed Up & Go test for ambulatory children with cerebral palsy. *Developmental Medicine & Child Neurology*, 57: 27.
Carmick, J. (2013). Forefoot Mobility in Ankle and Foot Orthoses: Effect on Gait of Children With Cerebral Palsy. *Pediatric Physical Therapy*, 25(3):331-337.
Centre for Reviews and Dissemination. (2009). *Systematic Reviews.* York: CRD, University of York.
Chan, B. C., Perkins, D., Wan, Q., Zwar, N., Daniel, C., Crookes, P., & Harris, M. F. (2010). Finding common ground? Evaluating an intervention to improve teamwork among primary health-care professionals. *International Journal for Quality in Health Care*, 22(6):519–524.
Chen, Y.-x., Yu, G.-r., Mei, J., Zhou, J.-q., & Wang, W. (2008). Assessment of subtalar joint neutral position: a cadaveric study. *Chinese Medical Journal*, 121(8):735-739.
Cans, C., Dolk, H., Platt, M. J., Colver, A., Prasauskiene, A., & Kragelogh-Mann, I., SCPE Collaborative Group. (2007). Recommendations from the SCPE collaborative group for defining and classifying cerebral palsy. *Developmental Medicine & Child Neurology Supplement*, 109:35-38.
Christovao, T., Pasini, H., Grecco, L., Ferreira, L., Duarte, N., & Oliveira, C. (2015). Effect of postual insoles on static and functional balance in children with cerebral palsy: A randomized controlled study. *Brazilian Journal of Physical Therapy*, 19(1): 44-51.
Clementz, B. G., & Magnusson, A. (1989). Assessment of tibial torsion employing fluroscopy, computed tomography and the cryosectioning technique. *Acta Radologica*, 30(1):75-80.
Craig, M. (1999). Physiotherapy management of cerebral palsy: current evidence and pilot analysis. *Physical Therapy Reviews*, 4(4):215–228.
Crenshaw, S., Herzog, R., Castagno, P., Richards, J., Miller, F., Michaloski, G., Moran, E. (2000). The Efficacy of Ton-Reducing Features in Orthotics on the Gait of Children with Spastic Diplegic Cerebral Palsy. *Journal of Pediatric Orthopaedics*, 20(2):210–216.
Dalvand, H., Dehghan, L., Feizi, A., Hosseini, S. A., & Armirsalari, S. (2013). The impacts of hinged and solid ankle-foot orthoses on standing and walking in children with spastic diplegia. *Iranian Journal of Child Neurology*, 7(4):12-19.
Danino, B., Erel, S., Kfir, M., Khamis, S., Batt, R., Hemo, Y., Hayek, S. (2015). Influence of orthosis on the foot progression angle in children with spastic cerebral palsy. *Gait & Posture*, 42(4):518-522.
Davis, E. (2013). *Custom foot orthotics for children*. Retrieved from Custon Foot Orthotics San Antonio: http://www.footorthotics.pro/customfootorthoticsforchildren.html
del Pilar Duque Orozco, M., Abousamra, O., Church, C., Lennon, N., Henley, J., Rogers, K. J., Miller, F. (2016). Reliability and validity of Edinburgh visual gait score as an evaluation tool for children with cerebral palsy. *Gait & Posture*, 49:14-18.

Dhote, S., Khatri, P., & Ganvir, S. (2012). Reliability of “Modified timed up and go” test in children with cerebral palsy. *Journal of Pediatric Neurosciences*, 7(2):96-100.

Dickens, W. S. (2006). Validation of a visual gait assessment scale for children with hemiplegic cerebral palsy. . *Gait & Posture*, 23(1):78–82.

Dickinson, H.O, Parkinson, K. N., Ravens-Sieberer, U., Schirripa, G., Thyen, U., Arnaud, C., Colver, A. F. (2007). Self-reported quality of life of 8–12-year-old children with cerebral palsy: a cross-sectional European study. *Lancet*, 369(9580): 2171-78.

Downs, S. H., & Black, N. (1998). The feasibility of creating a checklist for the assessment of the methodological quality both of randomised and non-randomised studies of health care interventions. *Journal of Epidemiology and Community Health*, 52(6):377-384.

Eckhoff, D. G., & Johnson, K. K. (1994). Three-dimensional computed tomographyreconstruction of tibial torsion. *Clinical Orthopaedics and Related Research*, 302:42-46.

Eddison, N., & Chockalingam, N. (2013). The effect of tuning ankle foot orthoses-footwear combination on the gait parameters of children with cerebral palsy. *Prosthetics and Orthotics International*, 37(2):95-107.

eorthopod.com. (2013). *External Tibial Torsion*. Retrieved from South Florida Institute of Sports Medicine: http://www.southfloridasportsmedicine.com/external-tibial-torsion.html

Evans, A. M., Rome, K., & Peet, L. (2012). The foot posture index, ankle lunge test, Beighton scale and the lower limb assessment score in healthy children: a reliability study. *Journal of Foot and Ankle Research*, 5(1):1.

Eyadah, A. A., & Ivanova, M. K. (2011). Methods for Measurement of Tibial Torsion. *The Kuwait Medical Journal*, 33(1):3-6.

Figueiredo, E. M., Ferreira, G. B., Moreira, R. C., Kirkwood, R. N., & Fetters, L. (2008). Efficacy of Ankle-Foot-Orthoses on Gait of Children with Cerebral Palsy: Systematic Review of Literature. *Pediatric Physical Therapy*, 20(3):207-223.

Fink, A. (2005). *How to conduct surveys: A step by step guide.* Thousand Oaks, CA: Sage.

Folinais, D., Thelen, P., Delin, C., Radier, C., Catonne, Y., Lazennec, J. Y. (2013). Measuring femoral and rotational alignment: EOS system versus computed tomography. *Orthopaedics & traumatology, surgery & research. Sep;99(5):509-516*.

Fosang, A. L., Galea, M. P., McCoy, A. T., Reddihough, D. S., & Story, I. (2003). Measures of muscle and joint performance in the lower limb of children with cerebral palsy. *Developmental Medicine and Child Neurology*, 45(10):664-670.

Gaumétou, E., Quijano, S., Ilharreborde, B., Presedo, A., Thoreux, P., Mazda, K., Skalli, W. (2014). EOS analysis of lower extremity segmental torsion in children and young adults. *Orthopaedics & traumatology, surgery & research. Sep;99(5):509-516*.

Gervasoni, E., Jonsdottir, J., Montesano, A., & Cattaneo, D. (2017). Minimal Clinically Important Difference of Berg Balance Scale in People With Multiple Sclerosis. *Archives of Physical Medicine and Rehabilitation*, 98(2):337-340.

Gibson, B. E. (2013, January). *The value of walking in children with CP: A matter of perception .* Retrieved from Lower Extremity Review: http://lermagazine.com/cover_story/the-value-of-walking-in-children-with-cp

Gigante, A., Bevilacqua, C., Bonetti, M. G., & Greco, F. (2003). Increased external tibial torsion in Osgood-Schlatter disease. *Acta Orthopaedica Scandinavica*, 74(4):431-436.

Golafshani, N. (2003). Understanding Reliability and Validity in Qualitative Research. *The Qualitative Report*, 8(4):597-606.

Gough, S., Yohannes, A. M., Thomas, C., & Sixsmith, J. (2013). Simulation-based education (SBE) within postgraduate emergency on-call physiotherapy in the United Kingdom. *Nurse Education Today*, 33(8):778-784.

Graham, KH., & Selber, P. (2003). Musculoskeletal Aspects of Cerebral Palsy. *The Journal of Bone & Joint Surgery (Br)*, 85(2):157-166.

Gupta, S., & Raja, K. (2012). Responsiveness of Edinburgh Visual Gait Score to Orthopedic Surgical Intervention of the Lower Limbs in Children with Cerebral Palsy. *American Journal of Physical Medicine & Rehabilitation*, 91(9):761-767.

Hainsworth, F., Harrison, M., Sheldon, T., & Roussounis, S. (2007). A preliminary evaluation of ankle orthoses in the management of children with cerebral palsy. *Developmental Medicine & Child Neurology*, 39(4):243-247.

Harradine, P., & Bevan, L. (2009). A review of the theoretical unified approach to podiatric biomechanics in relation to foot orthoses therapy. *Journal of American Podiatric Medical Association*, 99(4):317-325.

Harradine, P., Gates, L., & Bowen, C. (2018). If It Doesn't Work, Why Do We Still Do It? The Continuing Use of Subtalar Joint Neutral Theory in the Face of Overpowering Critical Research. *Journal of Orthopaedic & Sports Physical Therapy*, 48(3):130-132.

Harvey, A., & Gorter, J. (2011). Video gait analysis for ambulatory children with cerebral palsy: Why, when, where and how! *Gait & Posture*, 33(3):501-503.

Hicks, J., Arnold, A., Anderson, F., Schwartz, M., & Delp, S. (2007). The effect of excessive tibial torsion on the capacity of muscles to extend the hip and knee during single limb stance. *Gait & Posture*, 26(4):546-552.

Hootman, J. M., Driban, J. B., Sitler, M. R., Harris, K. P., & Cattano, N. M. (2011). Reliability and validity of three quality rating instruments for systematic reviews of observational studies. *Research Synthesis Methods*, 2(2):110-118.

Horner, B. (2000). The Effectiveness of Using Foot Orthotics as the Sole Intervention for the Treatment of Patellofemoral Pain Syndrome. *Masters Theses*, 626.

Houlihan, C. M., O'Donnell, M., & Conaway, R. D. (2004). Bodily pain and health-related quality of life in children with cerebral palsy. *Developmental Medicine & Child Neurology*, 46(5):305-310.

Howick, J., Chalmers, I., Glasziou, P., Greenhalgh, T., Heneghan, C., Liberati, A., . . . Thornton, H. (2011). *The 2011 Oxford CEBM Levels of Evidence (Introductory Document).* Retrieved from Oxford Centre for Evidence-Based Medicine.: https://www.cebm.net/index.aspx?o=5653

Hunter, S., & Burnett, G. (2000). Subtalar Joint Neutral and Orthotic Fitting. *Athletic Therapy Today*, Vol 5: 6-9.

Ibuki, A., Bach, T., Rogers, D., Bernhardt, J. (2010). An investigation of the Neurophysiologic Effect of Tone-Reducing AFOs on Reflex Excitability in Subjects with Spasticiy Following Stroke while Standing. *Prosthetics and Orthotics International*, 34(2):154–165.

Iatridou, G., & Dionyssiotis, Y. (2013). Reliability of balance evaluation in children with cerebral palsy. *Hippokratia*, 17(4):303-306.

Jagadamma, K., Coutts, F., Mercer, T., Herman, J., Yirrell, J., Forbes, L., van der Linden, M. (2015). Optimising the effects of rigid ankle foot orthoses on the gait of children with cerebral palsy (CP) - an exploratory trial. Disabil Rehabil Assist Technol. 2015;10. *Disabil Rehabil Assist Technol.*, 10(6):445-451.

Jahrling, L. (2001). Sensomotoric Orthotics. Germany.

Jakob, R. P., & Stussi, E. (1980). Tibial torsion cacluated by computed tomorgraphy and compared to other methods of measurement. *The Bone & Joint Journal*, 62-B(2):238-242.

Jarvis, H. (2013). *Investigation of the Podiatric Model of Foot Biomechanics.* (Unpublished doctoral dissertation, University of Salford, Salford United Kingdom). Retrieved from https://1library.net/document/zke1gkmz-investigation-of-the-podiatric-model-of-foot-biomechanics.html?utm_source=seo_title_list.

Jarvis, H., Nester, C., Bowden, P., & Jones, P. (2017). Challenging the foundations of the clinical model of foot function: further evidence that the root model assessments fail to appropriately classify foot function. *Journal of Foot and Ankle Research*, 10:7-18.

Jarvis, H., Nester, C., Jones, R., Williams A., & Bowden, P. (2012). Inter-assessor reliability of practice based biomechanical assessment of the foot and ankle. *Journal of Foot and Ankle Research*, 5:14.

Kane, K., Musselman, K., Manns, P., & Lanovaz, J. (2016). Toward evidence-informed AFO prescription: Identifying factors that guide clinician decision-making. Quebec City , PQ.

Kawamura, C., deMorais Filho, M., Barreto, M., dePaula Asa, S., Juliano, Y., & Novo, N. (2007). Comparison between visual and three-dimensional gait analysis in patients with spastic diplegic cerebral palsy. *Gait Posture*, 25(1):18-24.

Kembhavi, G., Darrah, J., Magill-Evans, J., & Loomis, J. (2002). Using the Berg Balance Scale to Distinguish Balance Abilities in Children with Cerebral Palsy. *Pediatric Physical Therapy*, 14(2):92-99.

Kennelly, J. (2011). Methodological Approach to Assessing the Evidence . *Reducing Racial/Ethnic Disparities in Reproductive and Perinatal Outcomes*, pp. 7-19 Springer.

Khermosh, O., Lior, G., & Weissman, S. L. (1971). Tibial Torsion in Children. *Clinical Orthopaedics*, 79: 25-31.

Knutson, L. M., & Clark, D. E. (1991). Orthotic devices for ambulation in children with cerebral palsy and myelomeningocele. *Physical Therapy*, 71(12):947–60.

Ko, J., & Kim, M. (2013). Reliability and Responsiveness of the Gross Motor Function Measure-88 in Children With Cerebral Palsy. *Physical Therapy*, 93(3):393-400.

Kooistra, B., Dijkman, B., Einhorn, T. A., & Bhandari, M. (2009). How to Design a Good Case Series. *The American Journal of Bone and Joint Surgery*, 91;Suppl 3:21-26.

Kristiansen, L. P., Gunderson, R. B., Steen, H., & Reikeras, O. (2001). The normal development of tibial torsion. *Skeletal Radiology*, 30:519-522.

Lee, S. H., Chung, C. Y., Park, M. S., Choi, I. H., & Cho, T.-J. (2009). Tibial Torsion in Cerebral Palsy: Validity and Reliability of Measurement. *Clinical Orthopaedics and Related Research*, 467(8):2098–2104.

Lee, J. H., Sung, I. Y., & Yoo, J. Y. (2008). Therapeutic effects of strengthening exercise on gait function of cerebral palsy. *Disability and rehabilitation*, *30*(19): 1439-1444.

Li, Y. H., & Leong, J. C. (1999). Intoeing gait in children. *Hong Kong Medical Journal*, 5(4):360-366.

Liodakis, E., Doxastaki, I., Chu, K., Krettek, C., Gaulke, R., Citak, M., & Kenawey, M. (2012). Reliability of the assessment of lower limb torsion using computed tomography: analysis of five different techniques. *Skeletal Radiology*, 41(3):305-311.

Liu, X., Kim, W., Dreup, B., & Mahadev, A. (2005). Tibial Torsion measurement by surface curvature. *Clinical Biomechanics*, 20(4):445-4450.

Ludwig, O., Kelm, J., Fröhlich, M. (2016). The influence of insoles with a peroneal pressure point on the electromyographic activity of tibialis anterior and peroneus longus during gait. *Journal of Foot and Ankle Research,* 9:33-41.

Ludwig, O., Quadflieg, R., & Koch, M. (2013). Einfluss einer sensomotorischen Einlage auf die Aktivität des M. peroneus longus in der Standphase. *Dtsch Z Sportmed*, 64:77-82.

Lundkvist Josenby, A., Jarnlo, G.-B., Gummesson, C., & Nordmark, E. (2009). Longitudinal Construct Validity of the GMFM-88 Total Score and Goal Total Score and the GMFM-66 Score in a 5-Year Follow-up Study. *Physical Therapy*, 89(4):342-350.

Lyons, K., Radburn, C., Orr, R., & Pope, R. (2017). A profile of injuries sustained by law enforcement officers: A critical review. *International Journal of Environmental Research and Public Health*, 14(2);142.

Mabuchi, A., Kitoh, H., Inoue, M., Hayashi, M., Ishiguro, N., & Suzuki, N. (2012). The Biomechanical Effect of the Sensomotor Insole on a Pediatric Intoeing Gait. *International Scholarly Network*. doi:10.5402/2012/396718

Martin, L., Baker, R., & Harvey, A. (2010). A Systematic Review of Common Physiotherapy Interventions in School-Aged Children with Cerebral Palsy. *Physical & Occupational Therapy in Pediatrics*, 30(4):294-312.

McBurney, H., Taylor, N. F., Dodd, K. J., & Graham, KH. (2003). A qualitative analysis of the benefits of strength training for young people with cerebral palsy. *Developmental Medicine & Child Neurology*, 45(10):658-663.

Menz, HB. (1995). Clinical hindfoot measurement; a critical review of the literature. *Foot*, 5:57-64.

Milner, C. E., & Soames, R. W. (1998). A comparison of four in vivo methods of measuring tibial torsion. *Journal of Anatomy*, 193(Pt 1):139-144.

Mohammed, F. M., Ali, S. M., & Mustafa, M. A. (2016). Quality of life of cerebral palsy patients and their caregivers: A cross sectional study in a rehabilitation center Khartoum-Sudan (2014 – 2015). *Journal of Neurosciences in Rural Practice*, 7(3):355-361.

Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PLoS Medicine*, 6 (7).

Morris, C. (2002). A review of the efficacy of lower-limb orthoses used for cerebral palsy. *Developmental Medicine & Child Neurology*, 44(3):205-211.

Morris, C., & Condie, D. (2009, September). Recent Developments in Healthcare for Cerebral Palsy. *Implications and Opportunities for Orthotics: Reports of a Meeting held at Wolfson College*. Oxford.

Morrison, S. C., & Ferrari, J. (2009). Inter-rater reliabiliay of the Foot Posture Index (FPI-6) in the assessment of the paediatric foot. *Journal of Foot and Ankle Research*, 2:26-31.

Muhamad, A. R., Freitas, J. M., Bomar, J. D., Dwek, J., Hosalkar. (2012). CT and MRI lower extremity torsional profile studies: measurement reproducibility. *Journal of Children's Orthopaedics Oct;6(5):391-396*.

Munro, B. H. (2004). *Statistical methods for health care research (5th edition).* Philadelphia: Lippincott Williams & Wilkins.

Murad, M., Sultan, S., Haffar, S., & Bazerbachi, F. (2018). Methodological quality and synthesis of case series and case reports. *BMJ Evidence-Based Medicine*, 23(2):60-63.

Najjarine, A. (n.d.). *Finding NCSP Using the NAS Anterior Lines Method.* icbmedical.com.

Najjarine, A., & Kielt, R. (2008). *The Orthotic Revolution; A Guide to Superior Biomechanics.* Sydney: The International College of Biomechanics.

Oeffinger, D. J., Tylkowski, C. M., Rayens, M. K., Davis, R. F., Gorton, G. E., D'Astous, J., . . . Luan, J. (2004). Gross Motor Function Classification System and outcome tools for assessing ambulatory cerebral palsy: a multicenter study. *Developmental Medicine and Child Neurology*, 46(5):311-319.

Oeffinger, D., Bagley, A., Rogers, S., Gorton, G., Kryscio, R., Abel, M., . . . Tylkowski, C. (2008). Outcome tools used for ambulatory children with cerebral palsy: responsiveness and minimum clinically important differences. *Developmental Medicine & Child Neurology*, 50(12):918-925.

Oeffinger, D., Gorton, G., Bagley, A., Nicholson, D., Barnes, D., Calmes, J., . . . Tylkowski, C. (2007). Outcome assessments in children with cerebral palsy, Part I: descriptive characteristics of GMFCS Levels I to III. *Developmental Medicine & Child Neurology*, 49(3):172-180.

Ohlendorf, D. (2013). Basics of Human Motion Control. *International Journal for Foot Orthotics: Foot & Shoe*, 1:19-24.

O'Brien, B. C., Harris, I. B., Beckman, T. J., Reed, D. A., & Cook, D. A. (2014). Standards for Reporting Qualitative Research: A Synthesis of Recommendations. *Academic Medicine*, 89(9):124-1251.

Ong, A., Hillman, S., & Robb, J. (2008). Reliability and validity of the Edinburgh Visual Gait Score for cerebral palsy when used by inexperienced observers. *Gait & Posture*, 28(2):323-326.

Owen, E. (2010). The importance of being earnest about shank and thigh kinematics especially when using ankle-foot orthoses. . *Prosthetics and Orthotics International*, 34(3):254–269.

Palisano, R., Rosenbaum, P., Walter, S., Russell, D., Wood, E., & Galuppi, B. (1997). Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Development Medicine & Child Neurology*, 39(4):214-23.

Patel, M. (2012). Tibial Torsion. *Medscape*.

Paulson, A., & Vargus-Adams, J. (2017). Overview of Four Functional Classification Systems Commonly Used in Cerebral Palsy. *Children (Basel)*, 4(4).

Perat, M. V. (2012). Rehabilitation of Children with Cerebral Palsy. *Eastern Journal Medicine*, 17:153-155.

Picciano, A., Rowlands, M., & Worrell, T. (1993). Reliability of open and closed kinetic chain subtalar joint neutral positions and navicular drop test. *Journal of Orthopaedic & Sports Physical Therapy*, 18:553-558.

Pierrynowski, M. R., & Smith, S. B. (1997). Effect of Patient Position on the Consistency of Placing the Rearfoot at Subtalar Neutral. *Journal of the American Podiatric Medical Association*, 87(9):399-406.

Piva, S. R., Fitzgerald, K., Irrgang, J. J., Jones, S., Hando, B. R., A, B. D., & Childs, J. D. (2006). Reliability of measures of impairments associated with patellofemoral pain syndrome. *BMC Musculoskeletal Disorders*, 7:33.

Putnam, R., Wening, J., & Hasso, D. (2014). Effectiveness of Edinburgh Visual Gait Score as a Valid Measure to Assess Influence of an Ankle Foot Orthosis on Adult Gait. (pp. February 26- March 1). American Academy of Orthotists & Prosthetists 40th Academy Annual Meeting & Scientific Symposium.

Radtka, S. A., Skinner, S. R., Dixon, D. M., & Johanson, M. E. (1997). Comparison of Gait With Solid, Dynamic, and No Ankle-Foot Orthoses in Children With Spastic Cerebral Palsy. *Physical Therapy*, 77(4):395-409.

Rathinam, C., Bateman, A., Peirson, J., & Skinner, J. (2014). Observational gait assessment tools in paediatrics – A systematic review. *Gait & Posture*, 40(2):279-285.

Read, H. S., Hazlewood, M. E., Hillman, S. J., Prescott, R. J., & Robb, J. E. (2003). Edinburgh Visual Gait Score for Use in Cerebral Palsy. *Journal of Pediatric Orthopaedics*, 23(3):296–301.

Rethlefsen, S., Blumstein, G., Kay, R., Dorey, F., & Wren, T. (2017). Prevalence of specific gait abnormalities in children with cerebral palsy revisited: influence of age, prior surgery, and Gross Motor Function Classification System level. *Developmental Medicine & Child Neurology*, 59(1):79-88.

Rethlefsen, S., Kay, R., Dennis, S., Forstein, M., & Tolo, V. (1999). The Effects of Fixed and Articulated Ankle-Foot Orthoses on Gait Patterns in Subjects with Cerebral Palsy. *Journal of Pediatric Orthopedics*, 19(4):470-474.

Ridgewell, E., Dobson, F., Bach, T., & Baker, R. (2010). A systematic review to determine best practice reporting guidelines for AFO interventions in studies involving children with cerebral palsy. *Prosthetics and Orthotics International*, 34(2):129-145.

Ries, A. J. (2017, January). Evaluating and Improving the Efficacy of Ankle Foot Orthoses for Children with Cerebral Palsy. *A Dissertation* . An Arbor, Michigan, United States of America: ProQuest LLC.

Ries, A. J., Novacheck, T. F., & Schwartz, M. H. (2015). The Efficacy of Ankle-Foot Orthoses on Improving the Gait of Children with Diplegic Cerebral Palsy: A Multiple Outcome Analysis. *American Academy of Physical Medicine and Rehabilitation*, 7(9):922-929.

Ritter, M. A., Derosa, G. P., & Babcock, J. L. (1976). Tibial Torsion? *Clinical Orthopaedics*, 120:159-163.

Robinson, L. W., Clement, N. D., Herman, J., & Gaston, M. S. (2017). The Edinburgh visual gait score – The minimal clinically important difference. *Gait & Posture*, 25-28.

Robinson, L. W., Clement, N., Fullarton, M., Richardson, A., Herman, J., Henderson, G., . . . Gaston, M. S. (2015). The relationship between the Edinburgh Visual Gait Score, the Gait Profile Score and GMFCS levels I-III. *Gait & Posture*, 741-743.

Romkes, J., & Brunner, R. (2002). Comparison of a dynamic and a hinged ankle-foot orthosis by gait analysis in patients with hemiplegic cerebral palsy. *Gait & Posture*, 15(1):18-24.

Root, M. L., Orien, W. P., & Weed, J. H. (1971). *Clinical Biomechanics: Normal and Abnormal Function of the Foot.* Los Angeles: Clinical Biomechanics Corp.

Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., Bax, M., Damaniano, D., … Jacobsson, B. (2007). A report: the definition and classification of cerebral palsy April 2006. *Developmental Medicine and Child Neurology. Supplement*, 109:8-14.

Russell, D., & Gorter, J. (2005). Assessing functional differences in gross motor skills in children with cerebral palsy who use an ambulatory aid or orthoses: can the GMFM-88 help? *Developmental Medicine & Child Neurology*, 47:462-467.

Russell, D., Rosenbaum, P., Cadman, D., Gowland, C., Hardy, S., & Jarvis, S. (1989). The Gross Motor Function Measure: a means to evaluate the effects of physical therapy. *Developmental Medicine & Child Neurology*, 31:341–352.

Sass, P., & Hassan, G. (2003). Lower extremity abnormalities in children. *American Family Physician*, 461-468.

Sell, K. E., Verity, T. M., Worrell, T. W., Pease, B. J., & Wigglesworth, J. (1994). Two Measurement Techniques for Assessing Subtalar Joint Position: A Reliability Study. *Journal of Orthopaedic & Sports Physical Therapy*, 162-197.

Shelly, A., Davis, E., Waters, E., Mackinnon, A., Reddihough, D., Boyd, R., . . . Graham, H. K. (2008). The relationship between quality of life and functioning for children with cerebral palsy. *Developmental Medicine & Child Neurology*, 50(3):199-203.

Shrout, P.E., Fleiss, J. L. (1979). Intraclass correlations: uses in assessing rater reliability. Psychological Bulletin, 86(2):420-428.

Smith, P., Hassani, S., Graf, A., Flanagan, A., Reiners, K., Kuo, K., . . . Harris, G. (2009). Brace Evaluation in Children with Diplegic Cerebral Palsy with a Jump Gait Pattern. *Journal of Bone and Joint Surgery*, 91(2):356-65.

Smith-Oricchio, K., & Harris, B. A. (1990). Interrater Reliability of Subtalar Neutral, Calcaneal Inversion and Eversion. *The Journal of Orthopaedic and Sports Physical Therapy*, 12:10-15.

Son, S. M., Ahn, S. H., Jung, G. S., Seo, S. W., Park, I. S., Song, J. C., . . . Do, K. H. (2014). The Therapeutic Effect of Tibia Counter Rotator With Toe-Out Gait Plate in the Treatment of Tibial Internal Torsion in Children. *Annals of Rehabilitation Medicine*, 38(2):218-225.

Song, D. H., Lee, Y., Eun, B. L., Lee, K. J., Kang, S. K., Vaq, S. G., . . . Kim, B. O. (2007). Usefulness of the tibia counter rotator (TCR) for the treatment of tibial internal torsion in children. *Korean Journal of Pediatrics*, 50(1):79-84.

Staheli, L. T., & Engel, G. M. (1972). Tibial Torsion. A Method of Assessment and a Survey of Normal Children. *Clinical Orthopaedics*, 86:183-186 .

Staheli, L. T., Corbett, M., Wyss, C., & King, H. (1985). Lower-extremity rotational problems in children. Normal values to guide management. *Journal of Bone & Joint Surgery*, 67(1):39-47.

Subke, J., Kolling, S., Griesemann, J., Kleinau, P., & Staudt, M. (2009). Optimization of Sensomotoric Insoles. *Article in IFMBE proceedings*.

Sullivan, G. M. (2011). A Primer on the Validity of Assessment Instruments. *Journal of Graduate Medical Education*, 3(2):119-120.

The KIDSCREEN Group. (2006). KIDSCREEN. Pabst Science Publishers.

Thomai, T., & Gita, R. (2017). Evaluation of the feasibility of the use of the Edinburgh Visual Gait Score (EVGS) for the assessment of gait in children with cerebral palsy. *International Journal of Therapy and Rehabilitation*, 1741-1645.

The Timed Up and Go. (2020). National Center for Injury Prevention and Control. *Centers for Disease Control and Prevention*. Website: https://www.cdc.gov/steadi/materials.html.

Uden, H., & Kumar, S. (2012). Non-surgical management of a pediatric "intoed" gait pattern – a systematic review of the current best evidence. *Journal of Multidisciplinary Healthcare*, 5:27-35.

Van Gheluwe, B., Kirby, K. A., Roosen, P., & Phillips, R. D. (2002). Reliability and Accuracy of Biomechanical Measurements of the Lower Extremities. *Journal of the American Podiatric Medical Association*, 92(6):317 - 326.

Varni, J. W., Burwinkle, T. M., Sherman, S. A., Hanna, K., Berrin, S. J., Malcarne, V. L., & Chambers, H. G. (2005). Health-related quality of life of children and adolescents with cerebral palsy: hearing the voices of the children. *Developmental Medicine & Child Neurology*, 47(9):592-597.

Viera, A. J., & Garrett, J. M. (2005). Understanding interobserver agreement: the kappa statistic. *Family Medicine*, 37(5):360-3.

Wegener, C., Wegener, K., Smith, R., Schott, K., & Burns, J. (2016). Biomechanical effects of sensorimotor orthoses in adults with Charcot–Marie– Tooth disease. *Prosthetics Orthotics International*, 40(4):436–446.

Westberry, D., Davids, J., Shaver, C., Tanner, S., Blackhurst, D., & Davis, R. (2007). Impact of Ankle-Foot Orthoses on Static Foot Alignment in Children with Cerebral Palsy. *American Journal of Bone & Joint Surgery*, 89(4):806-13.

White, H., Jenkins, J., Neace, W. P., Tylkowski, C., & Walker, J. (2002). Clinically prescribed orthoses demonstrate an increase in velocity of gait in children with cerebral palsy: a retrospective study. *Developmental Medicine & Child Neurology*, 44(4):227–232.

Williams, E. N., Carroll, S. G., Reddihough, D. S., Phillips, B. A., & Galea, M. P. (2005). Investigation of the timed 'Up & Go' test in children. *Developmental Medicine and Child Neurology*, 47(8):518-524.

Wingstrand, M., Hägglund, G., & Rodby-Bousquet, E. (2014). Ankle-foot orthoses in children with cerebral palsy: a cross sectional population based study of 2200 children. *BMC Musculoskeletal Disorders*, 15:327-33.

World Health Organization. (2006). Constitution of the World Health Organization. *Fifty-first World Health Assembly.* Basic Documents. Forty-fifth edition, Supplement.

Wren, T. A., Dryden, J. W., Mueske, N. M., Dennis, S. W., Healy, B. S., & Rethlefsen, S. A. (2015). Comparison of 2 Orthotics Approaches in Children with Cerebral Palsy. *Pediatric Physical Therapy*, 27(3):218-226.

Yam, W. K., & Leung, M. S. (2006). Interrater Reliability of Modified Ashworth Scale and Modified Tardieu Scale in Children With Spastic Cerebral Palsy. *Journal of Child Neurology*, 21(12):1031-1035.

Young, J., & Jackson, S. (2019). Improved motor function in a pre-ambulatory child with spastic bilateral cerebral palsy, using a custom rigid ankle-foot orthosis–footwear combination: A case report. *Prosthetics and Orthotics International*, 43(4):453–458.

Zhang, J., Wang, Y., Yang, Z., & Zhao, P. (2009). Comparison of the effects of dynamic and static ankle foot orthotics on children with spastic cerebral palsy. *Chinese Journal of Rehabilitation Medicine*, 24(1):45-48.

Appendices

a. Published Paper: Using the Edinburgh Visual Gait Score to Compare Ankle-Foot Orthoses, Sensorimotor Orthoses and Barefoot Gait Pattern in Children with Cerebral Palsy

children

MDPI

Article

Using the Edinburgh Visual Gait Score to Compare Ankle-Foot Orthoses, Sensorimotor Orthoses and Barefoot Gait Pattern in Children with Cerebral Palsy

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Abstract: Gait analysis is one aspect of evaluation in ambulatory children with cerebral palsy (CP). Ankle-foot orthoses (AFOs) improve gait and alignment through providing support. An alternative and under-researched orthosis are sensomotoric orthoses (SMotOs). The Edinburgh Visual Gait Score (EVGS) is a valid observational gait analysis scale to measure gait quality. The aim of this study was to use the EVGS to determine what effect AFOs and SMotOs have on gait in children with CP. The inclusion criteria were: mobilizing children with a CP diagnosis, no surgery in the past six weeks, and currently using SMotOs and AFOs. Eleven participants were videoed walking 5 m (any order) barefoot, in SMotOs and AFOs. Of the participants (age range 3–13 years, mean 5.5 ± 2.9), two were female and six used assistive devices. Seven could walk barefoot. Participants had spastic diplegia (4), spastic quadriplegia (6), and spastic dystonic quadriplegia (1). Gross Motor Functional Classification System (GMFCS) levels ranged I–IV. The total score for SMotOs (7.62) and AFOs (14.18) demonstrated improved gait when wearing SMotOs (no significant differences between barefoot and AFOs). SMotOs may be a viable option to improve gait in this population. Additional study is required but SMotOs may be useful in clinical settings.

Keywords: ankle-foot orthoses; sensorimotor orthoses; gait; Edinburgh Visual Gait Score; cerebral palsy; children

1. Introduction

Cerebral palsy (CP) is a neurodevelopmental condition well recognized to begin at birth or early childhood and persist through the lifespan [1]. It has been defined as a group of permanent disorders of the development of movement and postures, causing activity limitation through spasticity [2,3], muscle weakness, impaired postural control, and selective motor control as some of the primary manifestations of this brain injury [3]. CP is often accompanied by disturbances of sensation, perception, cognition, communication, behavior, and by epilepsy. One of these activity limitations may be the ability and co-ordination for walking (gait), with control of movements and postures being affected. As ambulation is the usual method for mobilizing, many children with CP strive to achieve any form of walking possible, whether it is with or without an assistive device.

Gait assessment assists in determining the degree and cause of gait abnormality and can be used as an outcome measure to evaluate change and the effectiveness of an intervention [4–6]. Instrumented gait analysis is the gold standard for the evaluation of movement [4] but requires highly technological equipment in a specialised gait laboratory. A gait laboratory requires considerable capital investment, trained personnel, and is often not readily accessible for routine clinical work [6,7]. Observational gait assessment is considered as a cost effective alternate for instrumented gait analysis in regular clinical practice [4]. One observational tool used in clinical settings is the Edinburgh Visual Gait Score (EVGS). The EVGS has been demonstrated as a valid and reliable [8,9], clinically applicable visual gait analyses tool for children with CP [9].

Ankle-foot orthotics (AFOs) are the typical prescription of lower extremity orthoses for the management of lower limb deformities that often occur with CP. A consensus conference of the International Society for Prosthetics and Orthotics identified the aims of lower extremity orthotic management in children with CP: (1) to correct and/or prevent deformity, (2) provide a base of support, (3) facilitate training of motor skills, and (4) improve efficiency of walking [10]. The goal of each AFO prescribed for a child with CP is the collective improvement of these biomechanical variables to increase the ease of taking an individual step with the potential to enhance walking activity and functional skills [11]. AFOs are designed to; affect body structure [3], support normal joint alignment and mechanics, provide variable range of motion (ROM) when appropriate, facilitate function [3,12–14], stabilize the ankle/foot complex [15], and enable a continuous Achilles/gastrocnemius stretch [16–18]. Along with joint alignment, other improvements that may be seen through the use of AFOs are an improvement in walking efficiency [10,19], position of the foot for function, [20], and improvement in gait function and pain prevention [21].

There are multiple studies assessing and comparing the motor changes in gait and other gross motor skills using different types of AFOs in children with CP [15,22,23]. Buckon et al. [15], noted that the AFOs did not significantly improve standing skills of the GMFM-88 ($p \leq 0.025$), but reported an improvement of the walking, running, and jumping elements when wearing AFOs ($p < 0.002$) compared to barefoot. In another study, Buckon et al. [22] noted that despite the AFO enhancing stability throughout static (e.g., standing) and dynamic (e.g., walking) gross motor and functional skills, they did not allow the child to achieve a skill they were previously unable to master. Dalvand et al. [23] also noted improvements in gait with both solid-AFO and hinged-AFO, although hinged-AFO demonstrated better improvements in gross motor function than solid-AFO.

Unlike AFOs, sensomotoric orthotics (SMotO) provide a different approach to the management of gait in children with CP. Wegner et al. [24] describe one adaptation theory as 'elements' on the foot orthoses (e.g., forefoot valgus posting or lateral rearfoot padding) increase local pressures which are detected by cutaneous receptors, muscle spindles or the Golgi apparatus. To expand this, SMotOs are created to directly change the muscle length [25] and "activate and deactivate" muscles by increasing or decreasing individually placed point specific pressure on musculotendinous structures in the foot of the tibialis posterior, peroneus brevis, and the lumbricals/quadratus plantae. This theory implies the information that is transmitted by the sensors for the control of muscle activity is changed [26]. Depending on these individual pressure bumps' height and placement, the muscles can be activated or restricted [25,26]. CP affects the different areas of the brain, therefore interrupting signals sent to the muscles. The SMotOs work on the idea that the signals are being sent from the muscles back up to the spinal cord through activation of the Golgi bodies, therefore signaling muscles to respond to stimulus. Clinically, the SMotO have demonstrated improved foot alignment, balance, control with walking, and functional skills. Clients have been prescribed the SMotOs as a supplementary lower limb orthosis when it is noted they are finding functional movement restricted in the AFO. SMotOs have also been prescribed for children who require more feedback from their feet, where wearing shoes alone has not been effective.

There is a lack of evidence examining the 'sensorimotor response' paradigm, as there, there are no randomized trials, minimal peer-reviewed papers in English, and only a few small cross-sectional pediatric papers [24]. There is only one paper in English reporting the use of SMotOs on in-toeing gait in children (with idiopathic in-toeing or clubfoot) and it found that they improved abnormal gait patterns of pediatric in-toeing gait by decreasing femoral internal rotation through the end of the swing phase and the beginning of the stance phase and by decreasing tibial internal rotation during the stance phase [27].

There are numerous papers that demonstrate improved gait when wearing AFOS, but there have been no studies to date comparing the effect of SMotOs and AFOs on gait in children with CP. Clinically, there appears to be improvements in gait quality when children with CP wear SMotOs. Therefore, this study aims to compare the changes in gait from barefoot when children with CP are wearing SMotOs and AFOs, through use of the EVGS.

2. Experimental Section

Ethical approval was obtained through the Bond University Research and Ethics Committee (Approval RO-1835). Consent was gained from clinic directors in both private practice settings. Participants and their caregivers were given an explanatory statement and consent form both of which were read and completed before data collection took place.

The participant inclusion criteria were: (a) child with a diagnosis of CP, (b) no surgery in past six weeks, (c) currently using SMotO and AFOs (completed the wearing in process of at least two weeks), and (d) able to mobilize (with or without a device). Participants were recruited by convenience sampling and were assessed through two private pediatric therapy practices in Sydney (Therapies for Kids and NAPA Centre). There was no limit on participation due to GMFCS level. Participants brought their own SMotOs and AFOs to the data collection sessions.

The AFOs were custom-made through public or private orthotists from polypropylene with velcro straps holding the foot in place. The AFOs all had a full-length foot plate. Due to the AFOs being the participants 'usual prescription' there was no further assessment or measurements of the AFOs performed. The SMotOs were custom made for each child from ethyl vinyl acetate and had been assessed and prescribed by a podiatrist or pedorthist who were both expert in this design type of orthosis. Figure 1 shows the finished SMotO [24]. Figure 2 illustrates the placement of toes (circles) metatarsal heads (crosses), and plantar fascia (lines).

Figure 1. Sensorimotor orthosis [24].

The Golgi bodies (in the musculotendinous junctions of the tendons of the foot) are activated to switch the muscle 'on' or 'off' by the pressure from the 'bumps' coinciding with the musculotendinous junctions [28] (Figure 2). It has been proposed that information is sent by afferent feedback pathways (centrally) in order to reduce the activity of over-active muscles through inhibition, which in turn facilitates an increase in the activity of weaker muscles [29]. Given the physiology of ascending muscle chains, the reaction will not only affect the single muscle targeted (e.g., in the foot) but influence the complete chain of movement (e.g., ankle and lower limb) and positively impact malposition [26] (e.g., over pronation).

The AFOs and SMotOs were worn inside the participants usual shoe (for that particular orthoses). Participants were asked to walk barefoot (where appropriate, due to ability to mobilize without any type of foot support), in SMotOs, and in AFOs for at least 5m at a self-directed pace. To provide motivation, participants were able to choose the order in which they wore the orthoses. Video imagery was taken with a handheld device (Apple iPhone 7s, Apple Inc., CA, USA) in anterior, posterior and lateral views. Videos were taken in a well-lit environment, following for lateral views or zooming in and out (as needed) when the child walked. If required due to poor video quality, misbehaviour, or misstep, additional videos were taken to ensure a quality video was assessed. The child was allowed the comfort of a break between walks if needed. To ensure validity and reliability of video, the plane of motion was followed.

Figure 2. Sensorimotor orthosis with descriptive markings.

To determine changes in gait, the EVGS was used. The EVGS comprises 17 parameters for each lower limb and evaluates movement across six sites (trunk, pelvis, hip, knee, ankle, and foot) [9]. Each gait phase is analyzed in the frontal, sagittal, and transverse planes and the anatomical sites are evaluated for movement through video observation [30]. Scoring uses a 3-point ordinal scale. When the segment is marked 0, it determines a normal score. When there is a 1, it means a moderate deviation from normal in either direction, and 2 relates to a marked deviation, therefore a higher score relates to a more severe deviation or abnormality of gait. The developers of EVGS reported a score reduction of 4 on each limb (compared to pre-intervention score) as an improvement and as the minimum change in score required that would be indicative of change, not merely related to observer variation [9].

The EVGS is a valid, robust, reliable, and easy-to-use observational gait analysis scale to measure gait quality in children with CP [4,9,31]. It has been examined for the purpose of orthosis evaluation in adults [32] but not yet validated in children with orthoses. The scale has stringent instructions to ensure reliability. Its agreement and validity with three-dimensional gait analysis have been documented [8] and was noted to be 52%–73%. The essential properties of an observation scale are validity, reliability, and ability to detect change [33]. Responsiveness is the ability of a tool's detection of change due to an intervention or over time. The EVGS is shown to correlate with the Gait Profile Score and the GMFCS [34], two relevant and valid measures relating to CP. Frame by frame analysis was performed to score the gait using the EVGS with all analysis performed by the principal researcher, a physiotherapist who works with the children and has 6 years of experience using this tool. This analysis took place after all face-to-face data collection had been completed, thereby minimizing time pressures on the families and their child. As the participant walked 5m for each camera direction, there was more than one stride to observe. Once viewed, the most usual score was used.

Data Analysis

The EVGS was analyzed through SPSS statistics software (Version 20.0, IBM, NY, USA) and the Microsoft Office Excel 2007 (Microsoft, WA, USA) were used for the data entry and analysis. Normality was determined via visual inspection of histograms, box plots and normal Q-Q plots. Depending on distribution, parametric or non-parametric tests were used to determine if there were any significant differences in the baseline characteristics or of the groups. Descriptive statistics were used to profile the data: the median difference of the total EVGS scores and the mean difference of the average walking score, before and after the intervention, were calculated. A summated score of each limb was used for data analysis for this study. Thus, the score for the EVGS ranges from 0 to 34 on left (L) or right (R). The data for the barefoot condition and both orthoses were then statistically analyzed through a one-way ANOVA to determine significance and post hoc Bonferroni to outline further comparison significance. There was also a cumulative total of each segment analysed. Repeated measures of ANOVA with Bonferroni post hoc and Wilk's Λ was performed. Alpha levels were set at 0.05 a priori.

3. Results

3.1. Participants

From 27 possible participants, 10 were unable to participate due to inability to understand instruction or poor comprehension, and four were unable to attend data collection. From the 13 potential participants, two were excluded as they did not wear AFOs anymore. See Table 1 for full participant demographics.

Table 1. Participant demographics.

Participant	CP Type	GMFCS Level	Age (whole Years)	Walking Aid
1	Sp Dip	III	3	Reverse walker
2	Sp Q	III	7	Reverse walker
3	Sp Dip	I	4	
4	Sp Dip	II	4	
5	Sp Q	IV	3	Rifton Pacer
6	Sp Q	II	6	
7	Sp Q	II	13	
8	Sp Dip	III	3	Reverse walker
9	Sp Dys Q	IV	4	Buddy Roamer
10	Sp Q	IV	6	Reverse walker
11	Sp Q	II	8	

CP = cerebral palsy; Sp Dip = spastic diplegia; Sp Q = spastic quadriplegia; Sp Dys Q = spastic dystonic quadriplegia; GMFCS = Gross Motor Function Classification System.

Of the final yield of 11 participants (aged between 3–13 years with average 5.5 ± 2.9 years), seven were able to walk barefoot and therefore had barefoot data collection recorded. Four were unable to walk barefoot due to inability or child refusal. There were four participants with spastic diplegia, six with spastic quadriplegia and one with spastic dystonic quadriplegia. The GMFCS levels of participants were: one level I, four level II, three level III, three level IV. Six participants used assistive devices for EVGS (participants 1, 2, 8, and 11 used a reverse walker, participant 6 used a Rifton pacer, and participant 10 used a Buddy Roamer). Five participants wore hinged-AFOs (participants 3, 4, 6, 7, and 11) and the remaining participants wore solid-AFOs. One participant had ethyl vinyl acetate (EVA) heel wedges on their solid-AFO to encourage weight through the heel, mimicking heel strike. There were no data recorded of orthosis use timing per child over usual day due to this research focusing on the immediate effect of orthoses on gait.

3.2. Scores

The total EVGS for (L) and (R) barefoot (where applicable) and with each orthosis are described in Table 2. See Appendix A for full extrapolated data set tables for each participant and segmental totals.

Table 2. Total (L) and (R) Edinburgh Visual Gait Score (EVGS) for participants.

Participant	GMFCS Level	Total score barefoot		Total score AFO		Total score SMotO	
		(L)	(R)	(L)	(R)	(L)	(R)
1	III	26	27	20	21	14	16
2	III	11	13	6	9	4	4
3	I	20	16	12	12	17	10
4	II	11	15	16	15	7	8
5	IV	13	9	12	11	8	8
6	II	25	26	14	16	7	10
7	II	12	14	8	9	4	5
8	III	-	-	18	20	11	14
9	IV	-	-	20	20	12	12
10	IV	-	-	14	14	6	6
11	II	-	-	14	11	4	5

GMFCS = Gross Motor Functional Classification Scale; (L) = left; (R) = right; AFO = ankle-foot orthoses; SMotO = sensomotoric orthoses; - = no data due to inability or refusal to walk barefoot.

When barefoot was assessed, the overall scores across participants demonstrated a poorer score than both AFO and SMotO, except for one participant who demonstrated poorer results when wearing AFOs compared to barefoot and SMotO. The descriptive statistics of EVGS are provided in Table 3 outlining the mean and SD for total (L) and (R) scores for barefoot, AFO, and SMotO intervention.

Table 3. Data descriptors for total (L) and (R).

Intervention		N	Mean	Std. Deviation
EVGS Total (L)	Barefoot	7	16.86	6.67
	AFO	11	14.00	4.47
	SMotO	11	8.55	4.43
Total (L)		29	12.62	5.94
EVGS Total (R)	Barefoot	7	17.14	6.77
	AFO	11	14.36	4.43
	SMotO	11	8.91	3.91
Total (R)		29	12.97	5.82

EVGS = Edinburgh Visual Gait Score; (L) = left; AFO = ankle-foot orthoses; SMotO = sensomotoric orthoses; (R) = right.

One-way ANOVA analyses revealed significant differences between total (L) ($p = 0.011$) and (R) ($p = 0.014$) scores between SMotO and AFOs (Table 4).

There were significant differences on the (L) lower limb between barefoot and SMotO ($p = 0.032$), and AFO and SMotO ($p = 0.027$). On the (R) lower limb, there were significant differences between AFO and SMotO ($p = 0.028$).

Table 4. Bonferroni comparison between the three orthoses.

	Intervention	Intervention	Significance
EVGS Total (L)	Barefoot	AFO	1.0
		SMotO	0.032 *
	AFO	Barefoot	1.0
		SMotO	0.027 *
EVGS Total (R)	Barefoot	AFO	1.0
		SMotO	0.052
	AFO	Barefoot	1.0
		SMotO	0.028 *

EVGS = Edinburgh Visual Gait Score; (L) = left; AFO = ankle-foot orthoses; SMotO = sensomotoric orthoses; (R) = right; * Indicates significance differences.

In the segmental analyses, repeated measures ANOVA elicited statistically significant differences in the foot, F (2, 5) = 8.993, $p < 0.022$; Wilk's $\Lambda = 0.218$, partial $\eta2 = 0.782$, and hip, F (2,5) = 6.10, $p < 0.045$; Wilk's $\Lambda = 0.290$, partial $\eta2 = 0.710$, with the biggest effect in the foot. Post hoc analysis with a Bonferroni adjustment revealed statistically significant differences between barefoot and SMotOs in the foot, mean difference (MD) = 8.86 (95% confidence interval [CI] 2.38 to 15.33, $p = 0.012$), and between AFO and SMotOs at the hip, MD = 1.14 (95% CI, 0.03 to 2.26, $p = 0.046$).

4. Discussion

The aim of this study was to investigate the changes two types of orthoses (SMotOs and AFOs) had on gait pattern in children with CP, as derived through the EVGS. All the participants were in GMFCS levels I–IV and used orthoses to walk, both in SMotOs and AFOs. There were six participants who required the use of ambulatory aids, displaying a varied range of gait ability. Overall, this cross-sectional cohort study found SMotOs to have a more positive influence on gait pattern compared to AFO and barefoot.

The total raw scores of each participant demonstrate that more desirable gait patterns were observed when wearing SMotO. This was resultant across 11 participants by a lower total score when wearing SMotOs on both the left (7.46) and right (7.77) compared to when wearing AFOs on the left (14.00) and right (14.36) and the seven participants with barefoot left (14.22) and right (14.11). Due to limitations in assessing calcaneal alignment in orthoses, the EVGS results would be affected at the foot / ankle when using restrictive orthoses compared to more dynamic orthoses that don't limit ankle movement. Barring participants 3 and 5, the general trend indicates that SMotO had the lower scores, which correlates to the EVGS score line indicating a more aligned gait pattern. The one-way ANOVA confirms that there was a significant difference in EVGS scores between the use of SMotO and AFO. One participant demonstrated a worse score on the (L) foot when wearing SMotO compared to AFO due to poor foot and knee alignment. Subsequently, a significant difference on the left lower limb was found when participants wore SMotOs compared to barefoot or AFOs, but not the right lower limb. The right lower extremity score was close to being significant ($p = 0.052$) when comparing SMotO to barefoot or AFOs but may need a larger yield study to determine its significance and if it is a usual trend. Interestingly, there was no significant difference between the AFO and barefoot scores.

Looking at the segmental breakdown of the EVGS, the hip and foot are seen to be most affected by orthotic intervention. It is found that that in the foot, barefoot is significantly different from SMotO ($p = 0.012$) and in the hip there is a significant difference between the AFO and SMotO ($p = 0.046$). The differences in the foot results between barefoot and SMotO may demonstrate the theory presented earlier by Wegner et al. [24]. Interestingly, there is no difference noted at the pelvis between barefoot and AFO. At the trunk, the AFOs presented a higher score than either barefoot or SMotOs, but this was not significant.

With regard to using the EVGS as an outcome measure to assess the effect of orthoses, a search for papers assessing the effect of AFOs on gait in children with CP through the EVGS only resulted in one case study paper by Young and Jackson [35]. This paper followed a child with spastic bilateral CP over 15 months, in which she was prescribed AFOs and began to stand and walk independently. Post AFO prescription, they noted clinically significant differences in the EVGS (increase by 7 points on the left and 11 points on the right—MCID of 2.4) and gait speed (42.9% increase in speed—MCID >10.9% is noted as large). This paper noted that AFOs did create a significant improvement in gait compared to barefoot. The lack of multiple papers using the EVGS to compare the effects of AFOs on gait in children with CP may provide a direction for future research.

One of the most common gait anomalies found in children with CP was in-toeing (amongst others) [36]. Although in a different population, there was one paper supporting the use of SMotOs to correct in-toeing in children with idiopathic in-toeing gait or clubfoot. Mabuchi [27] assessed the biomechanical effect of these orthoses on in-toeing gait in children. They found that the orthoses showed significant decreases in internal rotation at the proximal femur (loading response phase −18.3° ± 28.1° versus −21.6° ± 28.0°, $P = 0.009$ and terminal swing phase −16.3° ± 27.4° versus −19.0° ± 26.4°, $P = 0.047$) and the tibia in mid stance phase (0.7° ± 12.5° versus −2.0°± 14.9°, $P = 0.030$) and terminal stance phase (1.4° ± 11.9° versus −2.3° ± 14.5°, $P = 0.042$). They also found a significant increase in walking speed (67.9 m/min versus 64.9 m/min, $P < 0.001$) and stride length (500 mm versus 477 mm, $P < 0.001$). This may provide a basis to address in-toeing in children with CP with SMotOs.

The small, heterogenous sample affects the strength and generalizability of the results. Therefore, it is recommended that future research includes larger, more homogenous samples investigating SMotOs as another form of orthotic therapy for children with varied types of CP. It may also create opportunities to further investigate clinically useful observational gait assessment tools, such as the EVGS, for outcome measures when prescribing interventions such as orthoses. In support of Jagademma et al. [37], who stated that when investigating the effects of interventions such as AFOs, it is important to categorise children with CP based on their gait abnormalities. Therefore, it may be beneficial to further investigate multiple orthoses options or combinations than just AFOs alone, depending on the child's needs. Future research could include validating the EVGS as clinical assessment tool for use in children with lower limb orthoses, comparing customised, tuned AFOs with SMotOs in three-dimensional gait analysis, or expanding this study by removing limitations and performing over a longer time period.

Limitations of the Study: Due to the researchers only being able to draw participants from the population we had access to, there were limitations on the number of potential participants. Limitations to this research include a small final yield of participants available to collect the full range of data, mainly due to the restrictions uncovered such as comprehension of the task, ability to follow direction, and ability to attend data collection. This may affect the strength of the results, although papers within the literature demonstrate a range of participant numbers whilst using the EVGS from 7 [30] to 151 [31]. With a longer recruitment period and implementation of this as 'usual clinical practice' in prescription of SMotOs, a larger group could be assessed for continuation of these research findings. Another limitation was the lack of tuning the AFOs to avoid the possibility of iatrogenic gait compensations. Tuning of AFOs is recommended by Owen [38] and Eddison & Chockalingam [39] but was not performed in this setting as the researchers were investigating the usual prescription of AFOs and SMotOs on gait pattern. The angle in the ankle and knee in AFOs were not assessed for angle during this study which may provide a limitation to the strength of this study. Ideally, this study would have provided customised AFOs with in-depth explanation of prescription process. This would thus enhance the quality of study and reduce possible suboptimal AFO prescription. Unfortunately, this process is complex, can be expensive and was not possible at this stage. Potential bias is another limitation acknowledged in this paper and would be better resolved with EVGS completed by blinded raters with experience as well as more stringent patient preparation and video capture methodology. Along with this, the EVGS has not been assessed for reliability and validity when observing children in orthoses, possibly due to the inability to observe the calcaneus in orthoses or shoes. Ong, Hillman & Robb [40] validated the EVGS' reliability and validity for experienced observers in gait analysis. They noted, however, that the inexperienced observers were less accurate, and the experienced observers demonstrated more accurate results when compared to three-dimensional gait analysis. Three-dimensional gait analysis would be the preferred method of assessment for this type of study, but access to such a system was not possible. The EVGS does not allow for specific reporting of deviation direction, but rather indicated a deviation from 'normal', which may not be enough detail for some gait analyses.

5. Conclusions

The results of this study suggest that further evaluation of the effects of SMotO are warranted but the SMotO may, clinically, be an effective orthosis intervention to improve gait in children with CP. These results would be better validated if further research is performed in a gait laboratory using the gold standard three-dimensional gait analysis versus an observational score as there are many aspects of gait analysis. These results encourage further investigation into the use of SMotO in children with CP or to further specify the areas of benefit of the SMotO alongside AFO in relation to this population, their gait function and level of disability. Clinically, this creates an alternate orthoses prescription possibility for children with CP.

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Appendix A

Table A1. Participants with three scores (participant 1–7).

CHILD>	Participant 1					
Section	**Score Barefoot**		**Score AFO**		**Score SMotO**	
	Left	Right	Left	Right	Left	Right
Foot	12	12	7	7	5	7
Knee	7	7	5	5	3	3
Hip	2	3	2	3	2	2
Pelvis	2	2	2	2	1	1
Trunk	3	3	4	4	3	3
TOTAL	26	27	20	21	14	16
EVGS rating	severe	severe	moderate	moderate	Moderate	moderate

Table A1. *Cont.*

Participant 2						
Section	Score Barefoot		Score AFO		Score SMotO	
	Left	Right	Left	Right	Left	Right
Foot	6	6	1	3	1	1
Knee	1	2	1	1	1	1
Hip	2	2	2	2	2	2
Pelvis	1	2	1	2	0	0
Trunk	1	1	1	1	0	0
TOTAL	11	13	6	9	4	4
EVGS rating	mild	moderate	mild	mild	Mild	mild
Participant 3						
Section	Score Barefoot		Score AFO		Score SMotO	
	Left	Right	Left	Right	Left	Right
Foot	11	10	6	6	10	11
Knee	4	1	1	1	3	4
Hip	1	2	1	2	1	1
Pelvis	2	1	2	1	1	2
Trunk	2	2	2	2	2	2
TOTAL	20	16	12	12	17	10
EVGS rating	moderate	moderate	moderate	moderate	Moderate	mild
Participant 4						
Section	Score Barefoot		Score AFO		Score SMotO	
	Left	Right	Left	Right	Left	Right
Foot	5	6	7	7	2	2
Knee	3	3	3	3	2	1
Hip	1	2	2	1	0	1
Pelvis	2	2	2	2	1	2
Trunk	0	2	2	2	2	2
TOTAL	11	15	16	15	7	8
EVGS rating	Mild	Moderate	Moderate	Moderate	Mild	Mild
Participant 5						
Section	Score Barefoot		Score AFO		Score SMotO	
	Left	Right	Left	Right	Left	Right
Foot	6	5	5	4	2	2
Knee	4	1	3	3	2	2
Hip	1	1	2	2	1	1
Pelvis	1	1	1	1	2	2
Trunk	1	1	1	1	1	1
TOTAL	13	9	12	11	8	8
EVGS rating	Moderate	Mild	Moderate	Mild	Mild	Mild
Participant 6						
Section	Score Barefoot		Score AFO		Score SMotO	
	Left	Right	Left	Right	Left	Right
Foot	12	13	5	6	3	5
Knee	5	5	4	5	0	1
Hip	4	4	2	2	1	1
Pelvis	0	0	0	0	0	0
Trunk	4	4	3	3	3	3
TOTAL	25	26	14	16	7	10
EVGS rating	Severe	Severe	Moderate	Moderate	Mild	Mild

Table A1. *Cont.*

	Participant 7					
Section	Score Barefoot		Score AFO		Score SMotO	
	Left	Right	Left	Right	Left	Right
Foot	6	7	4	4	1	3
Knee	2	4	2	2	2	2
Hip	2	2	0	0	0	0
Pelvis	1	0	0	1	1	0
Trunk	1	1	2	2	0	0
TOTAL	12	14	8	9	4	5
EVGS rating	Moderate	Moderate	Mild	Mild	Mild	Mild

Table A2. Participants with two scores: AFO and SMotO (participant 8–11).

CHILD>	**Participant 8**			
Section	**Score AFO**		**Score SMotO**	
	Left	**Right**	**Left**	**Right**
Foot	7	9	5	7
Knee	5	5	5	5
Hip	0	0	0	0
Pelvis	4	4	1	2
Trunk	2	2	0	0
TOTAL	18	20	11	14
EVGS rating	Moderate	Moderate	Mild	Moderate
CHILD>	Participant 9			
Section	Score AFO		Score SMotO	
	Left	Right	Left	Right
Foot	6	7	7	7
Knee	5	4	2	2
Hip	2	2	0	0
Pelvis	4	4	2	2
Trunk	3	3	1	1
TOTAL	20	20	12	12
EVGS rating	Moderate	Moderate	Moderate	Moderate
CHILD>	Participant 10			
Section	Score AFO		Score SMotO	
	Left	Right	Left	Right
Foot	7	7	3	3
Knee	3	3	2	2
Hip	3	3	1	1
Pelvis	0	0	0	0
Trunk	1	1	0	0
TOTAL	14	14	6	6
EVGS rating	Moderate	Moderate	Mild	Mild
CHILD>	Participant 11			
Section	Score AFO		Score SMotO	
	Left	Right	Left	Right
Foot	6	4	1	2
Knee	3	2	1	1
Hip	2	2	1	1
Pelvis	0	0	0	0
Trunk	3	3	1	1
TOTAL	14	11	4	5
EVGS rating	Moderate	Mild	Mild	Mild

Table A1. *Cont.*

	Participant 7					
Section	Score Barefoot		Score AFO		Score SMotO	
	Left	Right	Left	Right	Left	Right
Foot	6	7	4	4	1	3
Knee	2	4	2	2	2	2
Hip	2	2	0	0	0	0
Pelvis	1	0	0	1	1	0
Trunk	1	1	2	2	0	0
TOTAL	12	14	8	9	4	5
EVGS rating	Moderate	Moderate	Mild	Mild	Mild	Mild

Table A2. Participants with two scores: AFO and SMotO (participant 8–11).

CHILD>	**Participant 8**			
Section	**Score AFO**		**Score SMotO**	
	Left	**Right**	**Left**	**Right**
Foot	7	9	5	7
Knee	5	5	5	5
Hip	0	0	0	0
Pelvis	4	4	1	2
Trunk	2	2	0	0
TOTAL	18	20	11	14
EVGS rating	Moderate	Moderate	Mild	Moderate
CHILD>	Participant 9			
Section	Score AFO		Score SMotO	
	Left	Right	Left	Right
Foot	6	7	7	7
Knee	5	4	2	2
Hip	2	2	0	0
Pelvis	4	4	2	2
Trunk	3	3	1	1
TOTAL	20	20	12	12
EVGS rating	Moderate	Moderate	Moderate	Moderate
CHILD>	Participant 10			
Section	Score AFO		Score SMotO	
	Left	Right	Left	Right
Foot	7	7	3	3
Knee	3	3	2	2
Hip	3	3	1	1
Pelvis	0	0	0	0
Trunk	1	1	0	0
TOTAL	14	14	6	6
EVGS rating	Moderate	Moderate	Mild	Mild
CHILD>	Participant 11			
Section	Score AFO		Score SMotO	
	Left	Right	Left	Right
Foot	6	4	1	2
Knee	3	2	1	1
Hip	2	2	1	1
Pelvis	0	0	0	0
Trunk	3	3	1	1
TOTAL	14	11	4	5
EVGS rating	Moderate	Mild	Mild	Mild

Table A3. Segmental total.

Foot Totals			
Participant	**Barefoot**	**AFO**	**SMotO**
1	24	14	12
2	12	4	2
3	21	12	21
4	11	14	4
5	11	9	4
6	25	11	8
7	13	8	4
Total	117	72	55
Knee Totals			
Participant	Barefoot	AFO	SMotO
1	14	10	6
2	3	2	2
3	5	2	7
4	6	6	3
5	5	6	4
6	10	9	1
7	6	4	4
Total	49	39	27
Hip Totals			
Participant	Barefoot	AFO	SMotO
1	5	5	4
2	4	4	4
3	3	3	2
4	3	3	1
5	2	4	2
6	8	4	2
7	4	0	0
Total	29	23	15
Pelvis Totals			
Participant	Barefoot	AFO	SMotO
1	4	4	2
2	3	3	0
3	3	3	3
4	4	4	3
5	2	2	4
6	0	0	0
7	1	1	1
Total	17	17	13
Trunk Totals			
Participant	Barefoot	AFO	SMotO
1	6	8	6
2	2	2	0
3	4	4	4
4	2	4	4
5	2	2	2
6	8	6	6
7	2	4	0
Total	26	30	22

References

1. Rosenbaum, P.; Paneth, N.; Leviton, A.; Goldstein, M.; Bax, M. A report: The definition and classification of cerebral palsy April 2006. *Wash. UCP Res. Educ. Found.* **2006**, *109*, 8–14.
2. Romkes, J.; Brunner, R. Comparison of a dynamic and a hinged ankle-foot orthosis by gait analysis in patients with hemiplegic cerebral palsy. *Gait Posture* **2002**, *15*, 18–24. [CrossRef]
3. Wingstrand, M.; Hägglund, G.; Rodby-Bousquet, E. Ankle-foot orthoses in children with cerebral palsy: A cross sectional population based study of 2200 children. *BMC Musculoskelet. Disord.* **2014**, *15*, 327–333. [CrossRef] [PubMed]
4. Rathinam, C.; Bateman, A.; Peirson, J.; Skinner, J. Observational gait assessment tools in paediatrics—A systematic review. *Gait Posture* **2014**, *40*, 279–285. [CrossRef]
5. Kawamura, C.M.; de Morais Filho, M.C.; Barreto, M.M.; de Paula Asa, S.K.; Juliano, Y.; Novo, N.F. Comparison between visual and three-dimensional gait analysis in patients with spastic diplegic cerebral palsy. *Gait Posture* **2007**, *25*, 18–24. [CrossRef]
6. Harvey, A.; Gorter, J.W. Video gait analysis for ambulatory children with cerebral palsy: Why, when, where and how! *Gait Posture* **2011**, *33*, 501–503. [CrossRef]
7. Dickens, W.E.; Smith, M.F. Validation of a visual gait assessment scale for children with hemiplegic cerebral palsy. *Gait Posture* **2006**, *23*, 78–82. [CrossRef]
8. Del Pilar Duque Orozco, M.; Abousamra, O.; Church, C.; Lennon, N.; Henley, J.; Rogers, K.J.; Sees, J.P.; Connor, J.; Miller, F. Reliability and validity of Edinburgh visual gait score as an evaluation tool for children with cerebral palsy. *Gait Posture* **2016**, *49*, 14–18. [CrossRef]
9. Read, H.S.; Hazlewood, M.E.; Hillman, S.J.; Prescott, R.J.; Robb, J.E. Edinburgh Visual Gait Score for Use in Cerebral Palsy. *J. Pediatr. Orthop.* **2003**, *23*, 296–301. [CrossRef]
10. Morris, C.; Condie, D. *Recent Developments in Healthcare for Cerebral Palsy: Implications and Opportunities for Orthotics*; Report of a Meeting held at Wolfson College: Oxford, UK, 2008.
11. Bjornson, K.; Zhou, C.; Fatone, S.; Orendurff, M.; Stevenson, R.; Rashid, S. The Effect of Ankle-Foot Orthoses on Community-Based Walking in Cerebral Palsy: A Clinical Pilot Study. *Pediatr. Phys. Ther.* **2016**, *28*, 179–186. [CrossRef]
12. Brodke, D.S.; Skinner, S.R.; Lamoreux, L.W.; Johanson, M.E.; St Helen, R.; Moan, S.A. Effects of ankle-foot orthoses on the Gait of Children. *J. Pediatr. Orthop.* **1989**, *9*, 702–708. [CrossRef] [PubMed]
13. Knutson, L.M.; Clark, D.E. Orthotic devices for ambulation in children with cerebral palsy and myelomeningocele. *Phys. Ther.* **1991**, *71*, 947–960. [CrossRef] [PubMed]
14. White, H.; Jenkins, J.; Neace, W.P.; Tylkowski, C.; Walker, J. Clinically prescribed orthoses demonstrate an increase in velocity of gait in children with cerebral palsy: A retrospective study. *Dev. Med. Child. Neurol.* **2002**, *44*, 227–232. [CrossRef] [PubMed]
15. Buckon, C.E.; Sienko Thomas, S.; Jakobson-Huston, S.; Moor, M.; Sussman, M.; Aiona, M. Comparison of three ankle–foot orthosis configurations for children with spastic diplegia. *Dev. Med. Child. Neurol.* **2004**, *46*, 590–598. [CrossRef] [PubMed]
16. Boyd, R.N.; Pliatsios, V.; Starr, R.; Wolfe, R.; Graham, H.K. Biomechanical transformation of the gastroc–soleus muscle with botulinum toxin A in children with cerebral palsy. *Dev. Med. Child. Neurol.* **2000**, *42*, 32–41. [CrossRef]
17. Morris, C. A review of the efficacy of lower-limb orthoses used for cerebral palsy. *Dev. Med. Child. Neurol.* **2002**, *44*, 205–211. [CrossRef]
18. Hainsworth, F.; Harrison, M.J.; Sheldon, T.A.; Roussounis, S.H. A preliminary evaluation of ankle orthoses in the management of children with cerebral palsy. *Dev. Med. Child. Neurol.* **2007**, *39*, 243–247. [CrossRef]
19. Rethlefsen, S.; Kay, R.; Dennis, S.; Forstein, M.; Tolo, V. The Effects of Fixed and Articulated Ankle-Foot Orthoses on Gait Patterns in Subjects with Cerebral Palsy. *J. Pediatr. Orthop.* **1999**, *19*, 470–474. [CrossRef]
20. Kane, K.; Musselman, K.; Manns, P.; Lanovaz, J. Toward evidence-informed AFO prescription: Identifying factors that guide clinician decision-making. In *Pedorthic Association of Canada Symposium*; PQ: Quebec City, QC, Canada, 2016.
21. Westberry, D.E.; Davids, J.R.; Shaver, C.; Tanner, S.L.; Blackhurst, D.W.; Davis, R.B. Impact of Ankle-Foot Orthoses on Static Foot Alignment in Children with Cerebral Palsy. *J. Bone Joint Surg. Am.* **2007**, *89*, 806–813. [CrossRef]

22. Buckon, C.E.; Sienko Thomas, S.; Jakobson-Huston, S.; Moor, M.; Sussman, M.; Aiona, M. Comparison of three ankle-foot orthosis configurations for children with spastic hemiplegia. *Dev. Med. Child. Neurol.* **2001**, *43*, 371–378. [CrossRef]
23. Dalvand, H.; Dehghan, L.; Feizi, A.; Hosseini, S.A.; Armirsalari, S. The impacts of hinged and solid ankle-foot orthoses on standing and walking in children with spastic diplegia. *Iran. J. Child. Neurol.* **2013**, *7*, 12–19. [PubMed]
24. Wegener, C.; Wegener, K.; Smith, R.; Schott, K.H.; Burns, J. Biomechanical effects of sensorimotor orthoses in adults with Charcot-Marie-Tooth disease. *Prosthet. Orthot. Int.* **2016**, *40*, 436–446. [CrossRef] [PubMed]
25. Subke, J.; Kolling, S.; Griesemann, J.; Kleinau, P.; Staudt, M. Optimization of sensomotoric insoles. In Proceedings of the World Congress on Medical Physics and Biomedical Engineering, Munich, Germany, 7–12 September 2009.
26. Ohlendorf, D. Basics of Human Motion Control. *Int. J. Foot Orthot. Foot, Shoe* **2013**, *1*, 19–24.
27. Mabuchi, A.; Kitoh, H.; Inoue, M.; Hayashi, M.; Ishiguro, N.; Suzuki, N. The biomechanical effect of the sensomotor insole on a pediatric intoeing gait. *ISRN Orthop.* **2012**, *2012*. [CrossRef] [PubMed]
28. Jahrling, L. Propriozeptiv oder „klassisch"? *Orthopädieschuhtechnik.* **2001**, *7*, 29–30.
29. Ludwig, O.; Quadflieg, R.; Koch, M. Einfluss einer sensomotorischen Einlage auf die Aktivität des M. peroneus longus in der Standphase. *Dtsch Z Sportmed.* **2013**, *64*, 77–82. [CrossRef]
30. Bella, G.P.; Rodrigues, N.B.; Valenciano, P.J.; Silva, L.M.; Souza, R.C. Correlation among the Visual Gait Assessment Scale, Edinburgh Visual Gait Scale and Observational Gait Scale in children with spastic diplegic cerebral palsy. *Rev. Bras. Fisioter.* **2012**, *16*, 130–140. [CrossRef]
31. Robinson, L.W.; Clement, N.D.; Herman, J.; Gaston, M.S. The Edinburgh visual gait score – The minimal clinically important difference. *Gait Posture* **2017**, *53*, 25–28. [CrossRef]
32. Putnam, R.; Wening, J.; Hasso, D. Effectiveness of Edinburgh visual gait score as a valid measure to assess influence of an ankle foot orthosis on adult gait. In Proceedings of the American Academy of Orthotists & Prosthetists 40th Academy Annual Meeting & Scientific Symposium, Chicago, IL, USA, 26 February–1 March 2014.
33. Gupta, S.; Raja, K. Responsiveness of Edinburgh Visual Gait Score to Orthopedic Surgical Intervention of the Lower Limbs in Children with Cerebral Palsy. *Am. J. Phys. Med. Rehabil.* **2012**, *91*, 761–767. [CrossRef]
34. Robinson, L.W.; Clement, N.; Fullarton, M.; Richardson, A.; Herman, J.; Henderson, G.; Robb, J.E.; Gaston, M.S. The relationship between the Edinburgh Visual Gait Score, the Gait Profile Score and GMFCS levels I-III. *Gait Posture* **2015**, *41*, 741–743. [CrossRef]
35. Young, J.; Jackson, S. Improved motor function in a pre-ambulatory child with spastic bilateral cerebral palsy, using a custom rigid ankle-foot orthosis–footwear combination: A case report. *Prosthet. Orthot. Int.* **2019**, *43*, 453–458. [CrossRef] [PubMed]
36. Rethlefsen, S.A.; Blumstein, G.; Kay, R.M.; Dorey, F.; Wren, T.A.L. Prevalence of specific gait abnormalities in children with cerebral palsy revisited: Influence of age, prior surgery, and Gross Motor Function Classification System level. *Dev. Med. Child. Neurol.* **2017**, *59*, 79–88. [CrossRef] [PubMed]
37. Jagadamma, K.C.; Coutts, F.J.; Mercer, T.H.; Herman, J.; Yirrell, J.; Forbes, L.; van der Linden, M.L. Optimising the effects of rigid ankle foot orthoses on the gait of children with cerebral palsy (CP)—An exploratory trial. *Disabil. Rehabil. Assist. Technol.* **2015**, *10*, 445–451. [CrossRef] [PubMed]
38. Owen, E. The importance of being earnest about shank and thigh kinematics especially when using ankle-foot orthoses. *Prosthet. Orthot. Int.* **2010**, *34*, 254–269. [CrossRef] [PubMed]
39. Eddison, N.; Chockalingam, N. The effect of tuning ankle foot orthoses-footwear combination on the gait parameters of children with cerebral palsy. *Prosthet. Orthot. Int.* **2013**, *37*, 95–107. [CrossRef] [PubMed]
40. Ong, A.M.L.; Hillman, S.J.; Robb, J.E. Reliability and validity of the Edinburgh Visual Gait Score for cerebral palsy when used by inexperienced observers. *Gait Posture* **2008**, *28*, 323–326. [CrossRef]

b. Published Paper: Sensomotoric Orthoses, Ankle–Foot Orthoses, and Children with Cerebral Palsy: The Bigger Picture

children

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Article

Sensomotoric Orthoses, Ankle–Foot Orthoses, and Children with Cerebral Palsy: The Bigger Picture

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Abstract: Ankle–foot orthoses (AFOs) and sensomotoric orthoses (SMotOs) are two—clinically relevant, yet under researched—types of lower limb orthoses used in children with cerebral palsy (CP). Quality of life is a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity. Evaluating the effect of these two types of orthoses on quality of life in children with CP has not been reported on. The aim of this case study series was to synthesise and enrich the volume of evidence reported to inform real world applications of SMotO use in children with CP. Participants recruited were children with CP who performed the Berg Balance Scale, Timed Up-and-Go, the Gross Motor Function Measure and/or the Edinburgh Visual Gait Score in AFOs, SMotOs and barefoot where able. Qualitative data included videos of gait, a questionnaire and pedographs. Eight participants completed 39 quantitative and six qualitative measures, with the Edinburgh Visual Gait Score (EVGS) reporting the highest response. A general improvement was seen in gross motor skills and gait when wearing the SMotOs compared to AFOs and some parents reported that SMotOs were preferred. The reader is able to correlate the quantitative results with the qualitative evidence presented.

Keywords: sensomotoric orthoses; ankle–foot orthoses; cerebral palsy; gait; gross motor skills; quality of life; children

1. Introduction

Ankle–foot orthoses (AFOs) and sensomotoric orthoses (SMotOs) are two types of lower limb orthoses. The benefits of using AFOs in children with cerebral palsy (CP) has been well documented over the years. AFOs are designed to affect body structure [1], support normal joint alignment and mechanics, provide variable range of motion (ROM) when appropriate, facilitate function [1–4], stabilise the ankle/foot complex [5] and enable a continuous Achilles/gastrocnemius stretch [6–8]. Along with joint alignment, other improvements that may be seen through the use of AFOs are improvements in walking efficiency [9,10], the position of the foot for function [11], and improvements in gait function and pain prevention [12]. Common types of AFOs seen in the literature are solid AFO (SAFO), hinged AFO (HAFO), and dynamic AFOs [5,13,14].

SMotO is a clinically relevant, yet under researched, orthoses option used in the same population. Unlike AFOs, SMotOs provide a different approach to the management of gait in children with CP. Wegner et al. [15], describe one adaptation theory as 'elements' on the foot orthoses (e.g., forefoot valgus posting or lateral rearfoot padding) increasing local pressures, which are detected by cutaneous receptors, muscle spindles or Golgi apparatus on musculotendinous structures in the foot of the tibialis posterior, peroneus brevis and the lumbricals/quadratus plantae. Depending on these individual pressure bumps' height and placement, the muscles can be activated or restricted [16,17]. CP affects the different areas of the brain, thereby interrupting signals sent to the muscles. The SMotOs work via the idea that the signals are being sent from the muscles back up to the spinal cord through activation of the Golgi bodies, therefore signalling muscles to respond to stimuli [15].

Quality of life (QoL) is defined by the World Health Organisation [18]: "health is a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity. The enjoyment of the highest attainable standard of health is one of the fundamental rights of every human being without distinction of race, religion, political belief, economic or social condition". With regard to CP, factors relating to QoL can include the child (age, gender, and severity of the disease; comorbidity and complications), family (socioeconomic status, relationships and support, coping mechanisms, parenting style, and knowledge about the disease) and the availability of management and rehabilitation services, as well as other environmental factors [19]. In a QoL study by Dickinson et al. [20], children with CP were investigated using KIDSCREEN (an instrument with scores in 10 domains) [21] and, through a comparison of the least and most able groups, severely limited self-mobility was significantly associated with a reduced mean score for physical wellbeing (7.6, 95% CI 2.7–12.4, $p = 0.002$), and pain was common and associated with a lower QoL in all domains. They concluded that physical impairments and presence of pain were responsible for variations (3% and 7%, respectively) in QoL. Therefore, a child's pain should be carefully assessed. When physical impairment impacts function, thus affecting QoL, therapists would likely make improving function a goal area for therapy. Independent walking is a typical goal of rehabilitation in children with CP, but this expectation can lead to frustration in parents and children, many of whom feel that they are more mobile and more functional when using assistive devices [22] versus completely independent.

Evaluating the effect of these two types of orthoses on quality of life in children with CP has not been reported on. Creating a 'real life' picture of particular 'cases' or participants in a mixed method case series study can bring depth to understanding both the clinical relevance and impact of an intervention on certain aspects of life. Although case series represent a low level of evidence (IV) and have methodological limitations with regard to making causal inferences about the relation between treatment and outcome [23], Murad et al. [24] suggested that when no other higher level of evidence is available, decision making can be informed using evidence derived from case reports and case series.

There is one published paper into the effect of SMotO on gait [25], and none on the effects of SMotO on gross motor skills and quality of life in children with CP. To provide a more complete picture of these complex children, a need to merge these studies in a select group of participants was found.

Therefore, noting the lack of literature in this field, the aim of this case study series was to synthesise and enrich the volume of evidence reported to inform real-world applications of SMotO use in children with CP. This case series also aims to demonstrate the impact of SMotOs and AFOs on function, movement and quality of life in the individual, in a way that is clinically relatable.

2. Materials and Methods

2.1. Study Design

Ethical process: Ethical approval was obtained through the Bond University Research and Ethics Committee (Approval RO-1835). Consent was gained from Clinic Directors in both private practice settings. Parents/caregivers were given an explanatory statement and consent form, both of which were read and completed before data collection took place. Consent was gained for video and image capture.

This study was a retrospective mixed method design, with a combination of both quantitative and qualitative outcome measures collected. Outcome measures were undertaken in two separate clinic locations, as well as six home settings due to the families being unable to travel. These settings were selected as they were familiar to the child and allowed the parents and/or siblings to be present throughout the testing. Relevant, pertinent participant qualitative information from the questionnaire (Q'AIRE) was extracted and combined with the correlating participants quantitative measurements to create the case series and is described in greater detail below.

2.2. Participants

Recruitment and inclusion criteria: Participants were children with CP recruited by convenience sampling through two private therapy practices (Therapies for Kids and NAPA Centre, Sydney, NSW, Australia). The inclusion criteria were: (a) diagnosis of CP with any Gross Motor Function Classification System (GMFCS) level, (b) using SMotOs/AFOs (or have used them) and completed the wearing in process, and (c) no surgery in past six weeks.

2.3. Intervention

All participants brought their own SMotOs and AFOs. The AFOs were all made from polypropylene with Velcro straps holding the foot in place. The SMotOs were custom made for each child from ethyl vinyl acetate (EVA). Each participant used SMotOs and/or AFOs whilst participating in outcome measures.

2.4. Quantitative Outcome Measures

The quantitative section of the case series process included the principal researcher assessing each participant as able using Timed Up-and-Go (TUG), the Berg Balance Scale (BBS), the Gross Motor Function Measure (GMFM-88), and/or the Edinburgh Visual Gait Score (EVGS). Each outcome measure was performed as the child was able, in any order deemed appropriate, and in any order of orthoses. For example, one child came in wearing AFOs and wanted to walk around the clinic; therefore, the EVGS in AFOs was assessed first. This child then became interested in some static activities; therefore, the GMFM in AFOs was performed next. Data collection continued for as many outcome measures as was possible for each of the participants' and within their ability and tolerance levels.

2.5. Qualitative Outcome Measures

Three styles of qualitative evidence were included: written feedback from parents compilated from the Q'AIRE, images of pedographs (pre- and post-SMotO) and/or video images of gait.

The Standards for Reporting Qualitative Research were followed [26]. A qualitative phenomenological approach was employed through a questionnaire-based survey. The Q'AIRE was designed to establish the effect of lower limb orthoses in current day to day QoL. This Q'AIRE was also undertaken to determine how the SMotO and AFO affect the child's function, as reported by parents. The Q'AIRE was emailed to multiple families after participating in quantitative data collection. Qualitative video images of a typical gait pattern were taken with the participant barefoot (where able), in AFOs and SMotOs. Video images were taken with a handheld device (Apple iPhone 7s, Apple Inc., Cupertino, CA, USA). The videos were taken in whichever location the outcome measures were recorded—either in clinic or at the participant's home—while the participant mobilised at a self-directed pace, using their usual prescribed walking aid (where necessary). Pedographs were supplied by the pedorthist.

3. Results

3.1. Participants and Outcome Measures

Data for eight participants (male: n = 7: female: n = 1) were collected. Participant 2 had EVA heel wedges on their SAFO to encourage weight through the heel, mimicking heel strike. The eight participants demonstrated a large range of physical abilities with reported GMFCS levels of I (n = 1 participant), II (n = 2 participants), III (n = 2 participants) and IV (n = 3 participants). The age range was three to 13 years (average age = 7 ± 3.7 years). Overall, there were 39 quantitative and six qualitative measures collected (Table 1). The EVGS demonstrated the highest response. Please note that, in videos, participants were previously coded (embedded in video) and, as such, may display a different participant number to the current number. The podiatrist and pedorthist who prescribed and fabricated the SMotOs provided pedograph images of two participants' footprints (7 and 8) before and after the use of SMotOs.

Table 1. Participant quantitative and qualitative outcome measure responses.

Outcome Measure		Intervention		
		SMotO	AFO	Barefoot
Quantitative	EVGS	7	6	2
	GMFM-88	5	5	0
	BBS	4	4	0
	TUG	3	3	0
		Responses		
Qualitative	Q'AIRE	4		
	Pedograph	2		
	Videography of gait	6		

SMotO: sensomotoric orthoses; AFO: ankle-foot orthoses; EVGS: Edinburgh Visual Gait Score; BBS: Berg Balance Scale; TUG: Timed Up-and-Go; Q'AIRE: Questionnaire.

3.2. Case Series

Data for each of the eight retained participants is presented below as individual cases.

Participant 1: Four-year-old male child with spastic diplegic CP, GMFCS III. The participant mobilises with a reverse walker. Participant 1 (Figure 1) demonstrated better scores in TUG, GMFM-88 and EVGS when in SMotO, likely due to the dynamic nature of the SMotOs being used in dynamic outcome measures (Table 2). Participant 1 displayed a better score in the BBS when in AFOs, likely due to the bracing effect of AFOs.

Figure 1. Participant 1.

Table 2. Participant 1 outcome measure comparative results between and sensomotoric orthoses (SMotOs) and ankle–foot orthoses (AFOs).

Outcome Measure	SMotO	AFO
TUG (s)	13.8 s	17 s
BBS (/56)	15	12
GMFM-88 (%)	73.51	71.17
EVGS	25 (total L & R)	38 (total L & R)

As per response from the Q'AIRE, the participant's mother reported that "I have been advised by some of our health care professionals that (my) son's gait is better in his AFOs than in Piedro (supportive disability shoe) with SMotO". This statement is contradicted by the EVGS results (Table 2). The mother of participant 1 did not give consent for video images of his gait.

Participant 2: Eight-year-old male child with spastic quadriplegic CP, GMFCS III. The participant mobilises with a reverse walker. Participant 2 (Figure 2) performed better in the TUG, GMFM-88 and EVGS when wearing SMotOs, likely due to the dynamic nature of the SMotOs being used in dynamic outcome measures (Table 3). Interestingly, the BBS reported the same score for both orthoses. Correlating videos (in DropBox folder link below) highlighting the participant's gait in SMotO, AFO and barefoot (as labelled) for 'Participant 2' have been provided for reference.

The participant's mother reported, as per the Q'AIRE, that "the SMotOs have been great for the stepping, sit to stand, pull to stand. Anything where he gets to feel the ground with the ankle movement has been the biggest bonus. Once I get some more supportive shoes to go with these then this will be the best. His Piedros still weren't helpful but we are looking at custom made ones to help this".

https://www.dropbox.com/sh/tfcrp9c1dxwmbn0/AAB9FSgGPunYpi8uCDDxZKpAa?dl=0

Figure 2. Participant 2.

Table 3. Participant 2 outcome measure comparative results between SMotO and AFO.

Outcome Measure	SMotO	AFO
TUG (s)	41.13	44.37
BBS (/56)	7	7
GMFM-88 (%)	69.11	64.41
EVGS	30 (total L & R)	41 (total L & R)

Participant 3: Four-year-old boy with spastic diplegic CP, GMFCS II. Participant 3 (Figure 3) demonstrated improved scores in the BBS, GMFM-88 and EVGS when wearing SMotOs compared to AFOs (Table 4). The GMFM-88 demonstrates a change of 6%, which is reported as a clinically important change in score. Correlating videos (in DropBox folder link below) highlighting the participant's gait in SMotO, AFO and barefoot (as labelled) for 'Participant 3' have been provided for reference.

The mother of Participant 3 reported, as per the Q'AIRE, that her "son is much more comfortable in SMotOs and finds it easier to manoeuvre his body and is much more willing to get up and try new things with them on because they're not as bulky".

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Figure 3. Participant 3.

Table 4. Participant 3 outcome measure comparative results between SMotO and AFO.

Outcome Measure	SMotO	AFO
TUG (s)	Unable to follow direction	
BBS (/56)	17	13
GMFM-88 (%)	85.51	79.51
EVGS	8 (total L & R)	15 (total L & R)

Participant 4: Thirteen-year-old girl with spastic quadriplegic CP, GMFCS II. Participant 4 (Figure 4) visually appeared to walk well in AFOs, but the results of the EVGS (Table 5) demonstrated a notable difference in the quality of her gait pattern when wearing AFOs compared to SMotO. Her GMFM-88 total score did not display a large difference in scores between orthoses, indicating that neither orthosis demonstrates an increased effect on gross motor skills compared to the other. Correlating videos (in DropBox folder link below) highlighting the participant's gait in SMotO, AFO and barefoot (as labelled) for 'Participant 4' have been provided for reference. Participant 4 did not complete the Q'AIRE.

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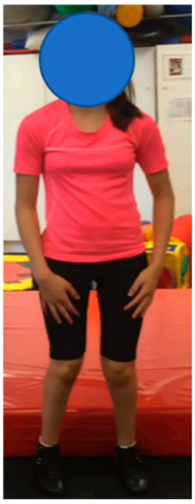

Figure 4. Participant 4.

Table 5. Participant 4 outcome measure comparative results between SMotO and AFO.

Outcome Measure	SMotO	AFO
TUG	11.33	10.13
BBS	39.00	37.00
GMFM-88 (%)	91.29	92.00
EVGS	15 (total L & R)	31 (total L & R)

Participant 5: Four-year-old boy with dystonic quadriplegic CP, GMFCS IV. The participant mobilises with a supportive reverse walker. Participant 5 (Figure 5) was physically affected by his dystonia and used a gait trainer to mobilise. He was unable to participate in any other outcome measures. Despite this limitation, the video images and EVGS both demonstrated the clear differences in his gait between barefoot, AFO and SMotOs. The qualitative evidence highlighting the participant's gait in the three conditions (correlating videos in DropBox folder link below) is supported by the results from the EVGS (Table 6).

https://www.dropbox.com/sh/8uolo7lhk5v5g2e/AAAV_tK_KujMt0Sew6t__p5ba?dl=0

Figure 5. Participant 5.

Table 6. Participant 5 comparative Edinburgh Visual Gait Score (EVGS) results between barefoot, AFO and SMotO.

Outcome Measure	Barefoot	AFO	SMotO
EVGS	51 (total L & R)	30 (total L & R)	17 (total L & R)

Participant 6: Five-year-old boy with spastic quadriplegic CP, GMFCS IV. The participant mobilises with a reverse walker and hip abduction brace. Participant 6 (Figure 6) struggled to walk without the support of his orthoses, walking frame and abduction brace. The results from the GMFM-88 (Table 7) showed a mild difference in scores between orthoses. Both the quantitative measure (EVGS) and qualitative images (video as per link below) demonstrated a difference in the quality of movement between orthoses and barefoot.

Correlating videos (in DropBox folder link below) highlighting the participant's gait in SMotO, AFO and barefoot (as labelled) for 'Participant 6' have been provided for reference. Participant 6 did not complete the Q'AIRE.

https://www.dropbox.com/sh/khmll6jwoumlpnv/AAAAgXhzhgX4PJGFpjRX6FR6a?dl=0

Figure 6. Participant 6.

Table 7. Participant 6 outcome measures comparative results between SMotO and AFO.

Outcome Measure	SMotO	AFO
GMFM-88 (%)	49.52	47.67
EVGS	9 (total L & R)	25 (total L & R)

Participant 7: Twelve-year-old boy with spastic dystonic quadriplegic CP, GMFCS I. Participant 7 (Figure 7) was independently mobile with and without shoes. Participant 7 demonstrated improved alignment and stability when he wore SMotO as per EVGS score (Table 8) compared to barefoot. The images from the pedographs (Figures 8 and 9) demonstrated weightbearing changes pre-SMotO and one year after using SMotO, especially through the right foot.

Correlating videos (in DropBox folder link below) highlighting the participant's gait in SMotO and barefoot (as labelled) for 'Participant 7' have been provided for reference. Participant 7 did not complete any other outcome measures.

https://www.dropbox.com/sh/i25vyb9jnd5v6f2/AABj5pBSJ4MNBj9kk4LPDbRSa?dl=0

Figure 7. Participant 7.

Table 8. Participant 7 EVGS results SMotO and barefoot.

Outcome Measure	SMotO	Barefoot
EVGS	3 (total L & R)	13 (total L & R)

Figure 8. Participant 7 left and right initial pedograph footprint (2016).

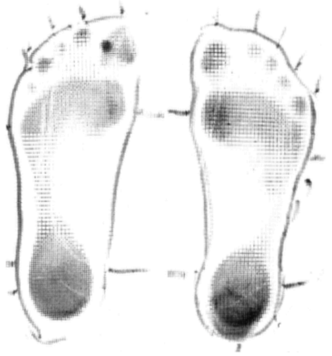

Figure 9. Participant 7 left and right final pedograph footprint (2017).

Participant 8: Six-year-old boy with dystonic quadriplegic CP, GMFCS IV. The participant mobilises with assistance in a supported walker. Participant 8 (Figure 10) was originally prescribed SAFOs then HAFOs despite not having any restriction in his ankle range of motion. He was able to bear weight with support and walks in a walker. He uses SMotOs in a Piedro shoe. Participant 8 was unable to complete any of the quantitative outcome measures due to his severe dystonia. From the Q'AIRE, mother reported "for children with CP—it appears there is a standard practice/framework for which children are expected to have/need. AFOs are one of these. I had to suggest my child transition from SAFO to HAFO. It was not suggested to us. They provide better support and ankle flexibility".

Figures 11 and 12 demonstrate the changes seen (over a seven-month period) in the muscle activation of his foot when wearing the SMotOs. These pedograph images corroborate the theory of the muscles learning to activate and support the foot, despite his CP diagnosis.

Figure 10. Participant 8.

Figure 11. Participant 8 left and right initial pedograph footprint (15 June 2016).

Figure 12. Participant 8 left and right final pedograph footprint (24 January 2017).

4. Discussion

Collecting a range of data in this population is challenging due to participants' age, level of disability, cognitive comprehension, inability to process instructions or give feedback, poor motor planning, and general behaviour. As such, data can often be incomplete when multiple outcome measures are collected. Given the challenges of collection and the volume and variety of data collected in this program of research, the use of a mixed method approach allowed for the collation of both quantitative and qualitive data to enrich the research findings. Furthermore, the inclusion of qualitative information to create a more holistic viewpoint of the intervention findings allowed the caregivers to validate and express their experiences.

The individual outcome measure results suggested a difference between areas of static and dynamic movement with SMotO and AFO. The general observed trend was increased static balance whilst wearing the AFO, and general improved ability (score) with dynamic movement when wearing the SMotO. This result is encouraging as a foundation to warrant further investigation into the use of SMotOs in this population for gross motor skills.

It appears, through both the qualitative and quantitative results, that children with CP have some preference for using SMotOs. In support of this, when looking at the Q'AIRE qualitative data, parents tended towards more positive comments regarding the use of SMotOs when compared to AFOs for gross motor skills and ease of use. In addition, it was identified that families do not have follow up appointments to reassess gait with AFOs and the impact of the AFO on gross motor skills. Clinically, it may also be beneficial to implement a follow-up timeline to reassess the effect of orthoses prescribed.

Using a clinic where a large number of clients reside interstate or internationally leaves the study somewhat lacking a significant number of potential participants, hence the low numbers of participants and inconsistent numbers of completed outcome measures. Some families had agreed to participate in all areas of the study but were unable to commit to the time required to complete the assessment, unable to participate due to distance or were unable to complete assessment due to the child's behaviour. A limiting factor of participants completing all outcome measures was the inability of the child to comprehend complex instructions relating to outcome measures, and participation was also affected due to participants' inability to process verbal instructions. However, the use of a gait aid did allow participants with lower functioning GMFCS levels to participate in the EVGS with individual results compared between orthoses. Another limitation is the bias towards the male gender, but future research could include a more even split between the genders.

Future Research Directions

While we acknowledge that this small case series demonstrates a minute part of the population, it does provide some important insights into the child as a 'whole picture' versus statistically based evidence alone. Future research could include creating a more specific assessment to determine the ability of the child, the family goals, and to investigate these in relation to the ability of SMotOs and AFOs to meet these goals, in order to create a more individualised, child-centred, goal-driven approach to orthoses prescription.

5. Conclusions

The aim of the current case series was to synthesise different participants' qualitative and quantitative evidence to support the volume of evidence in this thesis. It was also aimed at informing real-world applications of SMotO in children with CP using clinically relevant outcome measures relating to gait and gross motor skills orthoses prescription and what the best course of action for families and health professionals is.

It was concluded, based on the overall results of this case series, that a general improvement was seen in gross motor skills and gait when wearing the SMotOs compared to AFOs and they were preferred by some parents. By displaying video images of participants' gait in AFO, SMotO and barefoot, viewers are able to correlate quantitative results with the qualitative evidence of performance.

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References

1. Wingstrand, M.; Hägglund, G.; Rodby-Bousquet, E. Ankle-foot orthoses in children with cerebral palsy: A cross sectional population based study of 2200 children. *BMC Musculoskelet. Disord.* **2014**, *15*, 327–333. [CrossRef] [PubMed]
2. Brodke, D.S.; Skinner, S.R.; Lamoreux, L.W.; Johanson, M.E.; St. Helen, R.; Moran, S.A. Effects of ankle-foot orthoses on the Gait of Children. *J. Pediatr. Orthop.* **1989**, *9*, 702–708. [CrossRef] [PubMed]
3. Knutson, L.M.; Clark, D.E. Orthotic devices for ambulation in children with cerebral palsy and myelomeningocele. *Phys. Ther.* **1991**, *71*, 947–960. [CrossRef] [PubMed]
4. White, H.; Jenkins, J.; Neace, W.P.; Tylkowski, C.; Walker, J. Clinically prescribed orthoses demonstrate an increase in velocity of gait in children with cerebral palsy: A retrospective study. *Dev. Med. Child Neurol.* **2002**, *44*, 227–232. [CrossRef] [PubMed]
5. Buckon, C.; Sienko, T.; Jakobson-Huston, S.; Moor, M.; Sussman, M.; Aiona, M. Comparison of three ankle–foot orthosis configurations for children with spastic diplegia. *Dev. Med. Child Neurol.* **2004**, *6*, 590–598.
6. Morris, C. A review of the efficacy of lower-limb orthoses used for cerebral palsy. *Dev. Med. Child Neurol.* **2002**, *44*, 205–211. [CrossRef]
7. Boyd, R.N.; Pliatsios, V.; Starr, R.; Wolfe, R.; Kerr Graham, H. Biomechanical transformation of the gastroc–soleus muscle with botulinum toxin A in children with cerebral palsy. *Dev. Med. Child Neurol.* **2000**, *42*, 32–41. [CrossRef]
8. Hainsworth, F.; Harrison, M.; Sheldon, T.; Roussounis, S. A preliminary evaluation of ankle orthoses in the management of children with cerebral palsy. *Dev. Med. Child Neurol.* **2007**, *39*, 243–247. [CrossRef]
9. Morris, C.; Condie, D. Recent Developments in Healthcare for Cerebral Palsy. In Proceedings of the Implications and Opportunities for Orthotics: Reports of a Meeting held at Wolfson College, Oxford, UK, 8–11 September 2008.
10. Rethlefsen, S.; Kay, R.; Dennis, S.; Forstein, M.; Tolo, V. The Effects of Fixed and Articulated Ankle-Foot Orthoses on Gait Patterns in Subjects with Cerebral Palsy. *J. Pediatr. Orthop.* **1999**, *19*, 470–474. [CrossRef]
11. Kane, K.; Musselman, K.; Manns, P.; Lanovaz, J. *Toward Evidence-Informed AFO Prescription: Identifying Factors that Guide Clinician Decision-Making*; PQ: Quebec City, QC, Canada, 2016.
12. Westberry, D.; Davids, J.; Shaver, C.; Tanner, S.; Blackhurst, D.; Davis, R. Impact of Ankle-Foot Orthoses on Static Foot Alignment in Children with Cerebral Palsy. *J. Bone Joint Surg. Am.* **2007**, *89*, 806–813. [CrossRef]
13. Buckon, C.; Sienko, T.; Jakobson-Huston, S.; Moor, M.; Sussman, M.; Aiona, M. Comparison of three ankle-foot orthosis configurations for children with spastic hemiplegia. *Dev. Med. Child Neurol.* **2001**, *43*, 371–378. [CrossRef] [PubMed]
14. Dalvand, H.; Dehghan, L.; Feizi, A.; Hosseini, S.A.; Armirsalari, S. The impacts of hinged and solid ankle-foot orthoses on standing and walking in children with spastic diplegia. *Iran. J. Child Neurol.* **2013**, *7*, 12–19. [PubMed]
15. Wegener, C.; Wegener, K.; Smith, R.; Schott, K.; Burns, J. Biomechanical effects of sensorimotor orthoses in adults with Charcot–Marie– Tooth disease. *Prosthet. Orthot. Int.* **2016**, *40*, 436–446. [CrossRef] [PubMed]
16. Subke, J.; Kolling, S.; Griesemann, J.; Kleinau, P.; Staudt, M. Optimization of Sensomotoric Insoles. *Artic. IFMBE Proc.* **2009**, *25*, 554–557.
17. Ohlendorf, D. Basics of Human Motion Control. *Int. J. Foot Orthot. Foot Shoe* **2013**, *1*, 19–24.
18. World Health Organization. *Basic Documents: Forty-Ninth Edition (Including Amendments Adopted Up to 31 May 2019)*; World Health Organization: Geneva, Switzerland, 2020.
19. Mohammed, F.M.; Ali, S.M.; Mustafa, M.A. Quality of life of cerebral palsy patients and their caregivers: A cross sectional study in a rehabilitation center Khartoum-Sudan (2014–2015). *J. Neurosci. Rural Pract.* **2016**, *7*, 355–361. [CrossRef]
20. Dickinson, H.O.; Parkinson, K.N.; Ravens-Sieberer, U.; Schirripa, G.; Thyen, U.; Arnaud, C.; Beckung, E.; Fauconnier, J.; McManus, V.; Michelsen, S.I.; et al. Self-reported quality of life of 8–12-year-old children with cerebral palsy: A cross-sectional European study. *Lancet* **2007**, *369*, 2171–2178. [CrossRef]
21. The KIDSCREEN Group. *KIDSCREEN*; Pabst Science Publishers: Lengerich, Germany, 2006.

22. Gibson, B.E. The Value of Walking in Children with CP: A Matter of Perception. Available online: http://lermagazine.com/cover_story/the-value-of-walking-in-children-with-cp (accessed on 23 June 2018).
23. Kooistra, B.; Dijkman, B.; Einhorn, T.A.; Bhandari, M. How to Design a Good Case Series. *JBJS* **2009**, *91* (Suppl. S3), 21–26. [CrossRef]
24. Murad, M.; Sultan, S.; Haffar, S.; Bazerbachi, F. Methodological quality and synthesis of case series and case reports. *BMJ Evid. Based Med.* **2018**, *23*, 60–63. [CrossRef]
25. MacFarlane, C.; Hing, W.; Orr, R. Using the Edinburgh Visual Gait Score to Compare Ankle-Foot Orthoses, Sensorimotor Orthoses and Barefoot Gait Pattern in Children with Cerebral Palsy. *Children* **2020**, *7*, 54. [CrossRef]
26. O'Brien, B.C.; Harris, I.B.; Beckman, T.J.; Reed, D.A.; Cook, D.A. Standards for Reporting Qualitative Research: A Synthesis of Recommendations. *Acad. Med.* **2014**, *89*, 1245–1251. [CrossRef] [PubMed]

c. The Effect of Gait Plate Orthoses on Tibial Torsion in Children: A Retrospective Study

This study explores how orthoses are used to potentially correct lower limb alignment issues in children and if both these tools are a feasible option for use in later studies in children with cerebral palsy. This study was a part of the research journey; however, it was not included in the overall story of the thesis. With respect to cerebral palsy and tibial torsion, this is a contentious area, and as such the use of gait plates is unexplored.

Other concerns and considerations of this research were:

- Study design was a retrospective, non-controlled audit of clinical notes and presented with the aim to assess change in tibial torsion following gait plate intervention
- Variable age range and variable follow up (due to the nature of the patients' appointments and notes)
- The study investigated rotational changes with gait plate intervention within the tibia over time, but rotation is also expected to change with maturation, therefore, despite the results showing changes that were significantly increased to those in the literature, without a control group to compare to, the results add little to literature
- There was no MCID for malleolar position.

i. Introduction

Tibial torsion (TT) has been described as the twisting of the tibia about its longitudinal axis (Eyadah & Ivanova, 2011; Li & Leong, 1999; Liu, Kim, Dreup, & Mahadev, 2005; Milner & Soames, 1998; Patel, 2012). Torsion in the tibia is present throughout the lifetime, and a part of normal alignment that changes in the first few years of life (Staheli, Corbett, Wyss, & King, 1985). Torsion through the tibia is one of the factors thought to cause in-toeing (Son et al., 2014) and out-toeing in children (Sass & Hassan, 2003), whereby the child's foot turns to point excessively medially or laterally, respectively.

When the internal or external degree of torsion is excessive (Figure 18), it can affect lower limb alignment, biomechanics and gait of a person. Gigante, Bevilacqua, Bonetti, and Greco (2003) studied the relationship between Osgood-Schlatter disease and torsional abnormalities of the lower limb and found that increased external TT may play a role as a predisposing factor in the onset of Osgood-Schlatter disease in male athletes. In another study, Hicks, Arnold, Anderson, Schwartz, and Delp (2007) found that external TT deformity reduced the capacity of soleus to extend the knee during single limb stance. Excessive internal or external TT is thought to produce substantial abnormal pressure across the knee, particularly in those with cerebral palsy (CP) (Aiona, Calligeros, & Pierce, 2012). Aiona, Calligeros, and Pierce (2012) also found that correction of external TT post- distal internal rotation osteotomies improved the kinematic and kinetic deviations in ambulatory patients with CP.

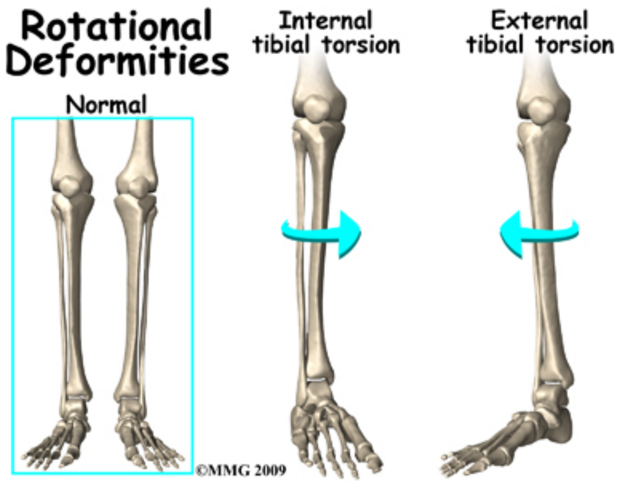

Figure 18: Rotational deformities of the tibia (eorthopod.com, 2013)

Physical examination is the most clinically relevant and affordable method of assessment. Li and Leong (1999) are in favour of physical examination over imaging as they note that sufficient information is obtained through a physical examination to formulate a treatment plan and do not usually require further imaging. Physical examination may be the best initial method of assessment for the presence of TT in a clinical setting (such as paediatric physiotherapy where parents may be hesitant to expose their child to extra radiation), but it is not the only method to determine TT. See Table 31 for other assessment tools used for the diagnosis and assessment of TT.

Table 31: Assessment tools for Tibial Torsion

Tools	Strengths	Weaknesses	Reference
Plain x-ray	Indicated when concerns with hips e.g., developmental dysplasia of the hip (DDH) or femoral anteversion	Not helpful in assessing TT	Li and Leong (1999)
Computed Tomography (CT)	Favoured by many authors as method to assess TT Noted by authors as a reference standard measure Best imaging technique for femoral anteversion, complex deformity or pre-surgery	Can be used for TT but clinical evaluation is generally sufficient	Butler-Manuel, Guy, and Heatley (1992); Clementz and Magnusson (1989); Eckhoff and Johnson (1994); Jakob and Stussi (1980); Lee et al. (2009); Liodakis et al. (2012)
EOS	The EOS system is a valid alternative to CT for lower-limb torsion measurement. EOS imaging allows a comprehensive evaluation in all three planes, such as for TT, while substantially decreasing patient radiation exposure	None reported	Folinais et al. (2013); Gaumétou et al. (2014)
Magnetic Resonance Imagery (MRI)	Magnetic resonance imaging (MRI) can be used as an alternative to CT, since there are many growing concerns of radiation exposure, particularly in the skeletally immature population	Anterior talus angle and posterior malleolar ankle are easier and more successful in determining TT	Basaran et al. (2015); Muhamad et al. (2012)
Ultrasonography	Can be used to measure the amount of femoral torsion or TT	Not as accurate as CT scan	Li and Leong (1999)
Gravity / Universal goniometer	Used to measure TT reliably	None reported relevant to TT	Bentley (2012); Piva et al. (2006); Song et al. (2007)

Knowledge of the normal range of TT at various ages is important in the assessment of the extent of a torsional deformity. Considering this, the literature reports varying ranges of torsion. Kristiansen, Gunderson, Steen, and Reikeras (2001) found that the average lateral torsion of the leg was 28° (at 4 years) with range of results 20°-37°, and that this increased at an average of 1° per year until ten years of age. After ten years old, the torsion was found to increase 4° until skeletal maturity, with a final mean lateral torsion of 38° (18°- 47°). Another study (Li & Leong, 1999) reported that during childhood the mean thigh foot angle = +10° with a range of -5° to +30°, which agrees with Khermosh, Lior, and Weissman (1971), Staheli and Engel (1972), and Ritter, Derosa, and Babcock (1976). These examples suggest that, throughout their lifetime, children progress through many degrees of rotation in their tibia before reaching skeletal maturity. The children who require intervention with gait plates (GPs) are those who have an in-toe or out-toe gait that affects their ability to safely mobilise or participate with other children. Figure 19 shows GPs to address internal and external TT.

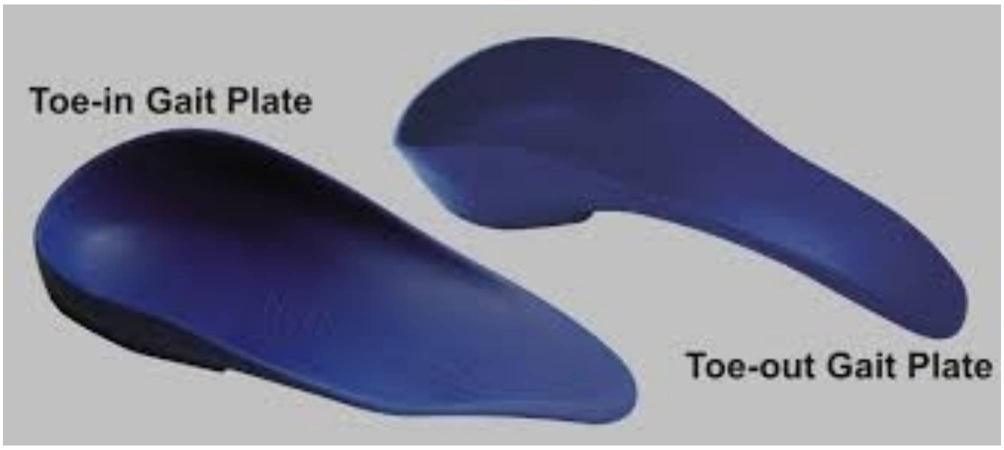

Figure 19: Orthotics with gait plate to address in-toe and out-toe gait (Davis, 2013)

The literature describes treatment such as surgery as a more extreme measure for more severe TT cases. Li and Leong (1999) found the only treatment options available for affected children were rotational osteotomy and observation. Staheli et al. (1985) stated long-term potential for disability with the absence of treatment should be weighed up against the risks of treatment and suggested that non-operative treatments were ineffective. These two options operate at extreme ends of treatment possibilities and may not appear feasible.

The treatment of TT with orthoses (and seeing the changes in torsion) gives rise to another option of treatment that is neither invasive nor inaction. The clinical and anecdotal evidence as given by a podiatrist suggests that using GPs to adjust the directional force where the children toe-off from (e.g., laterally or medially, depending on their torsion) will impact their torsional measures. However, no known research has been found to support this theory, and despite there being lack of evidence, researchers wanted to explore the relevance and applicability of the ALM and malleolar position (MP) to assess TT and if the orthoses changed TT. Malleolar position was used in this setting to assist to determine the presence of TT, by lining up the markings of the ALM in RCSP with the MP. It was hypothesised that the MP in typical children would demonstrate significant changes with the use of GPs. Therefore, the aim of this proof-of-concept study was to analyse the initial measurements of MP in children compared with the final measurements of MP post intervention, specifically application of orthoses with GP and report on changes in MP measurements. Depending on the results of this study, it will then serve to inform future papers and GPs as possible intervention in children with CP.

ii. Methods

Ethics approval was sought and approved (EC-00357). Clinical records were reviewed in this retrospective chart review study where the RCSP, NCSP and MP were assessed using the ALM technique, the MP specifically through using the lines on the anterior part of the foot. The ALM was previously determined to have very high inter- and intra- rater reliability with experienced assessors (ICC 0.96; 95% CI: 0.927-0.978 and ICC 0.96; 95% CI: 0.93 to 0.98) in adults, but only high in children if the assessor was highly experienced in the ALM technique. Therefore, only the clinical records were used from a highly experienced clinician.

Data inclusion criteria were: a) attending ICB Medical clinic, b) between ages two-18 years, c) receiving orthosis therapy for in- or out-toeing, d) attending more than one appointment where ALM/MP measurements were taken prior to orthosis prescription and post intervention. Exclusion criteria included: a) files that were incomplete (initial assessment date, date of treatment, date of birth, initial or subsequent measures), b) information was unable to be deciphered (poor handwriting, notes out of order), or c) the client attending only an initial appointment. Once all records meeting the intent of this study were collected, the following data were extracted; a) year of birth (from 1995 onwards - or as far back as the dates of appointments go for subjects who were under 18), b) the date of the initial consultation, c) initial and final RCSP in both left (L) and right (R), d) initial and final NCSP in both (L) and (R), e) initial and final MP in degrees in both (L) and (R) and f) the total amount of appointments. All the orthoses with GPs were assessed and prescribed using the same manner. The fabrication of all orthoses was performed by the same company in line with measurements by the clinician.

It was imperative the age was able to be extracted from the records to ensure only those under the age of 18 were included, as the study was aimed at assessing the influence of GP orthoses on changes in TT in a paediatric population. The date of the initial appointment and final / latest appointment was important as it gave a timeline per person, which could then be averaged with the number of appointments over the timeline of intervention. This allowed the calculation of the differences in torsion over time, and assessment of the effectiveness, or the change in measurement of TT with the intervention of GP orthoses to address either internal or external TT.

Once all data were collected, a descriptive analysis was performed. To determine change scores, scores from the final measure (T2) were subtracted from scores from the initial measure scores (T1) with the result recorded in their relative International System of Units (SI). To assess individual differences between measures, paired t-tests were performed. If a significant difference was found, then a linear regression was performed with change scores as the dependent variable and time period between measures as a covariate in order to determine the effect of time (and chronological aging) on change scores. Alpha levels were set at 0.05 *a priori*.

iii. Results

A total of 58 files were reviewed. Twenty-five charts were rejected due to meeting the exclusion criteria, resulting in a final yield of 33 files for review. There were 23 male participants and 10 female participants, and the initial age ranged from 2.5 - 14 years old (mean=6.03 ± 2.80). The mean period of time between T1 and T2 was 42.97 (±41.41; range 3 – 147) months. Descriptive results for all measures are shown below in Table 32.

Table 32: Descriptive table of measurements (°)

		MP(L)	MP(R)	NCSP(L)	NSCP(R)	RCSP(L)	RCSP(R)
Initial	**Mean T1**	-4.03	3.97	3.11	3.21	-9.79	-10.12
	SD T1	15.45	20.48	1.91	1.99	4.41	5.37
Final	**Mean T2**	9.30	12.88	3.5	3.5	-4.24	-4.12
	SD T2	8.77	10.39	1.35	1.35	2.75	2.88
Change	**Change**	13.33	8.91	0.39	0.29	5.55	6.00
	SD	-6.69	-10.10	-0.56	-0.64	-1.67	-2.49

MP=Malleolar Position; (L)=left; (R)=right; RCSP=Resting Calcaneal Stance Position; NCSP=Neutral Calcaneal Stance Position; SD=standard deviation; T1=initial measure; T2=final measure

Over the duration of the intervention period there was a significant change ($t(32)=-7.014$, $p<.001$) in RCSP (L) between the initial measure of initial appointment (T1) (mean = -9.79°±4.41°) and the follow up measure of final appointment (T2) (mean = -4.24°±2.75°) over a mean period of 42.97 (±41.41; range 3 – 147) months with a mean change in time of -5.94 (±5.78) months.

There was also a significant change ($t(32)=-6.279$, $p<.001$) in RCSP (R) between the initial measure of initial appointment (T1) (mean = -10.182°±5.32°) and the follow up measure of final appointment (T2) (mean = -4.12°±2.88°) over a mean period of 42.97 (±41.41; range 3 – 147) months with a mean change in time of -5.94 (±5.78) months.

A linear regression investigating contributing factors to change in the left leg noted that while time was not a significant contributor ($F(2,30)=29.21$, $p=.699$), the initial measure of (L) torsion was a significant contributor ($p<.001$). This was also found in the right leg with time not a significant contributor ($F(2,32)=42.73$. $p=.936$) and the initial measure of (R) torsion a significant contributor ($p<.001$).

Over the duration of the intervention, there was a significant change ($t(32)=-5.124$, $p<.001$) in MP (L) between the initial measure of initial appointment (T1) (mean=-4.03°±15.45°) and the follow up measure of final appointment (T2) (mean=9.30°±8.77°) over a mean period of 42.97 (±41.41; range 3 – 147) months with a mean change in time of -5.94 (±5.78) months.

There was also a significant change ($t(32)=-3.058$, $p<.004$) in MP (R) between the initial measure of initial appointment (T1) (mean = 3.97°±20.49°) and the follow up measure of final appointment (T2) (mean=12.88°±10.39°) over a mean period of 42.97 (±41.41; range 3–147) months with a mean change in time of -5.94 (±5.78) months.

As time was not a significant factor in the changes of (L) or (R) RCSP or MP, changes were likely due to the intervention or other unknown factors. However, a significant determinant of the amount of change was the initial (L) and (R) torsion score.

iv. Discussion

The aim of this study was to assess the effect of GP on TT over time. The hypothesis that there would be significant changes in the MP was confirmed. The results demonstrated a significant change ($p<.001$) in TT (RCSP and MP) over the course of treatment, with time not appearing to be a significant factor in these changes. These findings support the theory that changes in torsion may be due to the orthosis intervention and not necessarily normal developmental changes.

In the study by Son, et al., (2014), a tibial counter rotator (TCR) brace and / or a GP was implemented to effect a change in the internal TT. They report that both GP and TCR plus GP demonstrated significant ($p=0.001$) improvements in tibial alignment to neutral, however, the changes were more effective when both TCR and GP were worn. The results of this study align with the retrospective review, noting that GP can positively affect the torsion of the tibia when applied to internal TT. The retrospective data review, however, also found that applying an in-toeing GP to external TT was effective with correction to normal value.

The age of the participants ranged from 2.5 to 14 years at initial appointment, with the mean age of 6 years. Despite increases of TT being noted as 1^{o} per year from the age of 4 years (therefore after this age, the increase is of less clinical significance (Kristiansen et al, 2001), it was found that the average increase of TT was 13^{o}. It was noted that the effect of the GP orthoses was consistent, disregarding start age of orthosis use. This indicates that the effectiveness of these orthoses is relevant at the age it is applied, and that the general consensus of TT spontaneously correcting at seven years old may need to be revisited for those with excessive internal TT or external TT.

The results display a preliminary indication for use of gait plate orthoses that may be an option for intervention to correct excessive and abnormal TT in children. This intervention may be applied in a timelier manner rather than a 'wait and see' or surgical approach, which may cause excessive pain and recovery.

The children included were all actively weightbearing, with no data noted or recorded for children with significant disability, such as CP, that caused them to be predominately non-weightbearing. Ries (2017) noted that having stiff or contracted muscles, such as those with spastic CP, affected the ability of the bone growth and adversely affected the developing skeletal system due to the restricted or limited muscle force. The same author further reports that when the muscle force was abnormal, that bone growth may be abnormal and common deformities notes were excessive femoral anteversion, internal TT or external TT (Ries, 2017). Limitations include; weak design, low level evidence, no control group and small population sample. The effect of GPs on TT will need to be investigated by further research utilising a strong protocol and study design.

v. Conclusion

The results demonstrated that there were changes in the MP, which may be indicative of changes due to the GPs. As this was performed in a low level of research, as per the Oxford Centre for Evidence-Based Medicine Levels of Evidence (EBMLE), the results cannot be taken as gold standard. Unfortunately, due to the low level of evidence this paper served, the various limitations and poor results when assessing children (compared to adults), the ALM and MP assessment technique will not be used for future studies in this thesis. As there were no clear records of children with CP treated with GP orthoses, the effect of GPs in changing TT in children with CP was unable to be determined. This would be a direction for future studies where a control group can be used. The effect spasticity has on the skeletal system may indicate that this type of orthosis is only effective in those typically developing children who have active weightbearing through walking, whereby the torsional factor of the orthosis is the effector of change and is not negatively affected by spasticity. The children assessed in the reliability study and chart review were healthy (other than requiring orthotics for foot alignment) and had no disability, therefore this orthotic treatment style has not been investigated in children with CP.

d. Outcome Measures

i. Timed Up-and-Go (TUG)

The Timed Up and Go (TUG) was investigated by Williams, Carroll, Reddihough, Phillips, and Galea, (2005) in typical children and children with disabilities (spina bifida and CP) and found that it was responsive to change and can be used in children as young as three. This was agreed in the study by Iatridou and Dionyssiotis, (2013) who looked at three balance scales; TUG, Bruininks-Oseretsky Test of Motor Proficiency (BOTMP) and Berg Balance Scale (BBS) and reported these tests are considered to be reliable and valid tests, able to objectively define the quantitative mutation of the balance of the child in test with CP (Iatridou & Dionyssiotis, 2013).

The protocol for the TUG, employing the modifications described by Williams, et al., (2005) is detailed below:

1) Given a visual (concrete) target to reach compared to the more abstract instructions in the standard TUG
2) Instructions repeated as necessary throughout the test
3) An appropriately sized chair with or without arm rests (as necessary). This was further modified (for those children who were unable to stand up from sitting in their walker) to just walking on 'go'
4) Children were able to behave spontaneously, so no qualitative instructions (e.g., walk as fast as you can) were given to ensure naturalistic performance as indicative of their environment
5) Timing started when child stood or started to step (when already in standing) so it was valid for their movement

The TUG was scored in seconds by the assessor's stopwatch, with the final scores graded according to The Minimal Detectable Change (MDC) and Minimal Clinical Important Difference (MCID) for the TUG in children with CP, being: GMFCS Level I: 1.40 seconds, GMFCS Level II: 2.87 seconds, and GMFCS Level III: 8.74 seconds (Carey, Martin, Heathcock, & Comb-Miller, 2015).

Please see following for TUG assessment tool used along with the modifications as above.

Patient: ______________________ Date: ________ Time: ________ AM/PM

The Timed Up and Go (TUG) Test

Purpose: To assess mobility

Equipment: A stopwatch

Directions: Patients wear their regular footwear and can use a walking aid if needed. Begin by having the patient sit back in a standard arm chair and identify a line 3 meters or 10 feet away on the floor.

Instructions to the patient:

When I say **"Go,"** I want you to:

1. Stand up from the chair
2. Walk to the line on the floor at your normal pace
3. Turn
4. Walk back to the chair at your normal pace
5. Sit down again

On the word **"Go"** begin timing.

Stop timing after patient has sat back down and record.

Time: ________ seconds

An older adult who takes ≥12 seconds to complete the TUG is at high risk for falling.

Observe the patient's postural stability, gait, stride length, and sway.

Circle all that apply: ■ Slow tentative pace ■ Loss of balance ■ Short strides ■ Little or no arm swing ■ Steadying self on walls ■ Shuffling ■ En bloc turning ■ Not using assistive device properly

Notes:

For relevant articles, go to: **www.cdc.gov/injury/STEADI**

Centers for Disease Control and Prevention
National Center for Injury Prevention and Control

STEADI Stopping Elderly Accidents, Deaths & Injuries

ii. Berg Balance Scale (BBS)

The Berg Balance Scale (BBS) was initially intended for use in the elderly population with balance impairments or those with acute stroke (Berg et al,1992). The BBS was investigated by Iatridou and Dionyssiotis (2013) as a part of a three-balance test evaluation to determine the reliability in children with CP. Their investigation showed a low coefficient of variation amongst their comparative measures (mean 0.10 ± 0.32 SD).

This fourteen section test covers balance challenges a child may face in day-to-day activity (e.g., static standing without holding on, getting up and down from a chair, picking an object up off the floor). Each section has a five-point rating scale from 0-4, (0 = lowest level of function, 4 = highest level of function). The total score is calculated by adding all section scores, highest possible score being 56. The results, in raw scores, are then subject to the following interpretation: 41-56 = low fall risk, 21-40 = medium fall risk, 0 –20 = high fall risk. The minimal clinically important difference (MCID) for the BBS was noted by Gervasoni et al., (2017) as three points of difference.

Please see following for full scale used.

Berg Balance Scale

Scoring: Mark the lowest category that applies for each function.

Recommended for ACL of 4.0 or greater

	Date	Date	Date	Date
Sitting to Standing: *(Instructions: "Please stand up. Try not to use your hand for support."*) 4 = able to stand, no hands, and stabilizes independently 3 = able to stand independently using hands 2 = able to stand using hands after several tries 1 = needs minimal assist to stand or to stabilize 0 = needs moderate or maximal assist to stand				
Standing Unsupported: (*Instructions: "Stand for two minutes without holding.")* 4 = able to stand safely 2 minutes 3 = able to stand 2 minutes with supervision 2 = able to stand 30 seconds unsupported 1 = needs several tries to stand 30 seconds unsupported 0 = unable to stand 30 seconds unassisted				
Sitting Unsupported Feet On Floor: (*Instructions: "Sit with arms folded for two minutes.")* 4 = able to sit safely and securely for 2 minutes 3 = able to sit 2 minutes under supervision 2 = able to sit 30 seconds 1 = able to site 10 seconds 0 = unable to sit without support for 10 seconds				
Standing to Sitting (*Instructions: "Please sit down.")* 4 = sits safely with no or minimal use of hands 3 = controls descent by using hands 2 = uses back of legs against chair to control descent 1 = sits independently but has uncontrolled descent 0 = needs assistance to sit				
Transfers: (*Instruction: "Please move from chair to char/mat and back again. One way toward a seat with armrests and one way toward a seat without armrests.")* 4 = able to transfer safely with minor use of hands 3 = able to transfer safely, definitely need use of hands 2 = able to transfer with verbal cueing and/or supervision 1 = needs one person to assist 0 = needs two people to assist or supervise to be safe				
Standing Unsupported with Eyes Closed: (*Instructions: "Close your eyes and stand still for 10 seconds.")* 4 = able to stand 10 seconds safely 3 = able to stand 10 seconds with supervision 2 = able to stand 3 seconds 1 = able to stand for less than 3 seconds 0 = needs help to keep from falling				
Standing Unsupported with Feet Together: (*Instructions: "Place your feet together and stand without holding.")* 4 = able to place feet together independently and stand 1 minute safely 3 = able to place feet together independently and stand 1 min. with supervision 2 = able to place feet together independently but unable to hold for 30 seconds 1 = needs help to attain position but able to stand 15 sec. with feet together 0 = needs help to attain position and unable to hold for 15 seconds				

56 – Maximum Score
> 45 – less likely to fall
< 45 – more likely to fall

49.9 - 51.1 –	Needs no assistive device
47 - 49.6 –	Use of cane needed for outdoors
44 – 46.5 –	Use of can needed indoors as well as outdoors
26.7 – 39.6 –	Needs to use walker at all times

iii. Gross Motor Function Measure (GMFM-88)

The Gross Motor Function Measure (GMFM-88) was created for children with CP and was evaluated for its reliability and responsiveness in children with CP by Ko and Kim (2013) who validated it as excellent tool. It was also assessed in a study by Russell and Gorter (2005) who determined that the GMFM-88 was sensitive to within child changes in function, whether they used an ambulatory aid or orthoses or not. In a systematic review by Alotaibi, Long, Kennedy, & Bavishi, (2014), found that the GMFM-88 and GMFM-66 are both useful and of value when assessing children under the age of 17 in their gross motor skills. To enhance the knowledge of a child's functional ability, they also recommend clinicians to use other outcome measures in conjunction with the GMFM-88.

The GMFM-88 reflects the motor developmental sequence from birth to 5 years, it includes activities that precede or that are prerequisites for the achievement of gait (Dalvand, Dehghan, Feizi, Hosseini, & Armirsalari, 2013). The GMFM-88 was performed by the principal researcher as per the outlines in the GMFM-88 handbook and completed as per assessment sheet below. The guidelines for use as per CanChild website (https://canchild.ca/en/resources/44-gross-motor-function-measure-gmfm) describe a change of 5-7 percentage points as a medium positive change.

Characteristics of the Gross Motor Function Measure (GMFM-88) (Lundkvist Josenby, Jarnlo, Gummesson, & Nordmark, 2009)

Characteristic	GMFM-88
Purpose	Evaluation and determination of gross motor function capacity (descriptive, discriminative, predictive, and evaluative)
Target group	Children with cerebral palsy; validated also for Down syndrome and used in children with osteogenesis imperfecta and acute lymphatic leukemia
Equipment	Common physical therapy tools or equipment defined in the manual and score sheet
Administration	Clinical observation
Estimated test time required	60−45 min
Items	88
Scoring of single items	Ordinal 4-point scale for each item: 0=does not initiate task, 1=initiates task, 2=partially completes task, 3=completes task, NT=not tested; 3 trials allowed
Scoring of items that participants could not perform or that were not tested	0 points
Dimensions	Gross motor skills based on milestones in 5 dimensions: A, lying and rolling (17 items); B, sitting (20 items); C, kneeling and crawling (14 items); D, standing (13 items); and E, walking, running, and jumping (24 items)
Scale scoring	Dimension score: percentage of accomplished tasks in each dimension (A—E) Total score: mean of 5-dimension scores Goal total score: individualized for each child and including only dimensions of selected

	goal areas, calculated as mean of included dimension scores
Expected development	Available in tables according to age and Gross Motor Function Classification System (GMFCS) level in manual; all items can be accomplished by a 5-year-old child showing normal development
Interpretation tools	None

Please see below for the GMFM-88 assessment tool used.

GROSS MOTOR FUNCTION MEASURE (GMFM)
SCORE SHEET (GMFM-88 and GMFM-66 scoring)

Child's Name: ______________________ ID#: ______________

Assessment Date: ______________ GMFCS Level[1]:
year / month / day

☐ I ☐ II ☐ III ☐ IV ☐ V

Date of Birth: ______________
year / month / day

Chronological Age: ______________ Evaluator's Name: ______________
year / month / day

Testing Condition (e.g., room, clothing, time, others present):

____

The GMFM is a standardized observational instrument designed and validated to measure change in gross motor function over time in children with cerebral palsy. The scoring key is meant to be a general guideline. However, most of the items have specific descriptors for each score. It is imperative that the guidelines contained in the manual be used for scoring each item.

SCORING KEY
0 = does not initiate
1 = initiates
2 = partially completes
3 = completes
9 (or leave blank) = not tested (NT) [used for the GMAE-2 scoring*]

It is important to differentiate a true score of "0" (child does not initiate) from an item which is Not Tested (NT) if you are interested in using the GMFM-66 Ability Estimator (GMAE) Software.

*The GMAE-2 software is available for downloading from www.canchild.ca for those who have purchased the GMFM manual. The GMFM-66 is only valid for use with children who have cerebral palsy.

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CanChild
Centre for Childhood Disability Research

[1]GMFCS level is a rating of severity of motor function. Definitions for the GMFCS-E&R (expanded & revised) are found in Palisano et al. (2008). Developmental Medicine & Child Neurology. 50:744-750 and in the GMAE-2 scoring software. http://motorgrowth.canchild.ca/en/GMFCS/resources/GMFCS-ER.pdf

Check (3) **the appropriate score:** if an item is not tested (NT), circle the item number on the right column

	Item	A: LYING & ROLLING	SCORE				NT
	1.	SUP, HEAD IN MIDLINE: TURNS HEAD WITH EXTREMITIES SYMMETRICAL	0☐	1☐	2☐	3☐	1.
*	2.	SUP: BRINGS HANDS TO MIDLINE, FINGERS ONE WITH THE OTHER	0☐	1☐	2☐	3☐	2.
	3.	SUP: LIFTS HEAD 45°	0☐	1☐	2☐	3☐	3.
	4.	SUP: FLEXES R HIP & KNEE THROUGH FULL RANGE	0☐	1☐	2☐	3☐	4.
	5.	SUP: FLEXES L HIP & KNEE THROUGH FULL RANGE	0☐	1☐	2☐	3☐	5.
*	6.	SUP: REACHES OUT WITH R ARM, HAND CROSSES MIDLINE TOWARD TOY	0☐	1☐	2☐	3☐	6.
*	7.	SUP: REACHES OUT WITH L ARM, HAND CROSSES MIDLINE TOWARD TOY	0☐	1☐	2☐	3☐	7.
	8.	SUP: ROLLS TO PR OVER R SIDE	0☐	1☐	2☐	3☐	8.
	9.	SUP: ROLLS TO PR OVER L SIDE	0☐	1☐	2☐	3☐	9.
*	10.	PR: LIFTS HEAD UPRIGHT	0☐	1☐	2☐	3☐	10.
	11.	PR ON FOREARMS: LIFTS HEAD UPRIGHT, ELBOWS EXT., CHEST RAISED	0☐	1☐	2☐	3☐	11.
	12.	PR ON FOREARMS: WEIGHT ON R FOREARM, FULLY EXTENDS OPPOSITE ARM FORWARD	0☐	1☐	2☐	3☐	12.
	13.	PR ON FOREARMS: WEIGHT ON L FOREARM, FULLY EXTENDS OPPOSITE ARM FORWARD	0☐	1☐	2☐	3☐	13.
	14.	PR: ROLLS TO SUP OVER R SIDE	0☐	1☐	2☐	3☐	14.
	15.	PR: ROLLS TO SUP OVER L SIDE	0☐	1☐	2☐	3☐	15.
	16.	PR: PIVOTS TO R 90° USING EXTREMITIES	0☐	1☐	2☐	3☐	16.
	17.	PR: PIVOTS TO L 90° USING EXTREMITIES	0☐	1☐	2☐	3☐	17.
		TOTAL DIMENSION A					

	Item	B: SITTING	SCORE				NT
*	18.	SUP, HANDS GRASPED BY EXAMINER: PULLS SELF TO SITTING WITH HEAD CONTROL	0☐	1☐	2☐	3☐	18.
	19.	SUP: ROLLS TO R SIDE, ATTAINS SITTING	0☐	1☐	2☐	3☐	19.
	20.	SUP: ROLLS TO L SIDE, ATTAINS SITTING	0☐	1☐	2☐	3☐	20.
*	21.	SIT ON MAT, SUPPORTED AT THORAX BY THERAPIST: LIFTS HEAD UPRIGHT, MAINTAINS 3 SECONDS	0☐	1☐	2☐	3☐	21.
*	22.	SIT ON MAT, SUPPORTED AT THORAX BY THERAPIST: LIFTS HEAD MIDLINE, MAINTAINS 10 SECONDS	0☐	1☐	2☐	3☐	22.
*	23.	SIT ON MAT, ARM(S) PROPPING: MAINTAINS, 5 SECONDS	0☐	1☐	2☐	3☐	23.
*	24.	SIT ON MAT: MAINTAIN, ARMS FREE, 3 SECONDS	0☐	1☐	2☐	3☐	24.
*	25.	SIT ON MAT WITH SMALL TOY IN FRONT: LEANS FORWARD, TOUCHESTOY, RE-ERECTS WITHOUT ARM PROPPING	0☐	1☐	2☐	3☐	25.
*	26.	SIT ON MAT: TOUCHES TOY PLACED 45° BEHIND CHILD'S R SIDE, RETURNS TO START	0☐	1☐	2☐	3☐	26.
*	27.	SIT ON MAT: TOUCHES TOY PLACED 45° BEHIND CHILD'S L SIDE, RETURNS TO START	0☐	1☐	2☐	3☐	27.
	28.	R SIDE SIT: MAINTAINS, ARMS FREE, 5 SECONDS	0☐	1☐	2☐	3☐	28.
	29.	L SIDE SIT: MAINTAINS, ARMS FREE, 5 SECONDS	0☐	1☐	2☐	3☐	29.
*	30.	SIT ON MAT: LOWERS TO PR WITH CONTROL	0☐	1☐	2☐	3☐	30.
*	31.	SIT ON MAT WITH FEET IN FRONT: ATTAINS 4 POINT OVER R SIDE	0☐	1☐	2☐	3☐	31.
*	32.	SIT ON MAT WITH FEET IN FRONT: ATTAINS 4 POINT OVER L SIDE	0☐	1☐	2☐	3☐	32.
	33.	SIT ON MAT: PIVOTS 90°, WITHOUT ARMS ASSISTING	0☐	1☐	2☐	3☐	33.
*	34.	SIT ON BENCH: MAINTAINS, ARMS AND FEET FREE, 10 SECONDS	0☐	1☐	2☐	3☐	34.
*	35.	STD: ATTAINS SIT ON SMALL BENCH	0☐	1☐	2☐	3☐	35.
*	36.	ON THE FLOOR: ATTAINS SIT ON SMALL BENCH	0☐	1☐	2☐	3☐	36.
*	37.	ON THE FLOOR: ATTAINS SIT ON LARGE BENCH	0☐	1☐	2☐	3☐	37.
		TOTAL DIMENSION B					

	Item	C: CRAWLING & KNEELING	SCORE				NT
	38.	PR: CREEPS FORWARD 1.8m (6')	0☐	1☐	2☐	3☐	38.
*	39.	4 POINT: MAINTAINS, WEIGHT ON HANDS AND KNEES, 10 SECONDS	0☐	1☐	2☐	3☐	39.
*	40.	4 POINT: ATTAINS SIT ARMS FREE	0☐	1☐	2☐	3☐	40.
*	41.	PR: ATTAINS 4 POINT, WEIGHT ON HANDS AND KNEES	0☐	1☐	2☐	3☐	41.
*	42.	4 POINT: REACHES FORWARD WITH R ARM, HAND ABOVE SHOULDER LEVEL	0☐	1☐	2☐	3☐	42.
*	43.	4 POINT: REACHES FORWARD WITH L ARM, HAND ABOVE SHOULDER LEVEL	0☐	1☐	2☐	3☐	43.
*	44.	4 POINT: CRAWLS OR HITCHES FORWARD 1.8m(6')	0☐	1☐	2☐	3☐	44.
*	45.	4 POINT: CRAWLS RECIPROCALLY FORWARD 1.8m (6')	0☐	1☐	2☐	3☐	45.
*	46.	4 POINT: CRAWLS UP 4 STEPS ON HANDS AND KNEES/FEET	0☐	1☐	2☐	3☐	46.
	47.	4 POINT: CRAWLS BACKWARDS DOWN 4 STEPS ON HANDS AND KNEES/FEET	0☐	1☐	2☐	3☐	47.
*	48.	SIT ON MAT: ATTAINS HIGH KN USING ARMS, MAINTAINS, ARMS FREE, 10 SECONDS	0☐	1☐	2☐	3☐	48.
	49.	HIGH KN: ATTAINS HALF KN ON R KNEE USING ARMS, MAINTAINS, ARMS FREE, 10 SECONDS	0☐	1☐	2☐	3☐	49.
	50.	HIGH KN: ATTAINS HALF KN ON L KNEE USING ARMS, MAINTAINS, ARMS FREE, 10 SECONDS	0☐	1☐	2☐	3☐	50.
*	51.	HIGH KN: KN WALKS FORWARD 10 STEPS, ARMS FREE	0☐	1☐	2☐	3☐	51.

TOTAL DIMENSION C ☐

	Item	D: STANDING	SCORE				NT
*	52.	ON THE FLOOR: PULLS TO STD AT LARGE BENCH	0☐	1☐	2☐	3☐	52.
*	53.	STD: MAINTAINS, ARMS FREE, 3 SECONDS	0☐	1☐	2☐	3☐	53.
*	54.	STD: HOLDING ON TO LARGE BENCH WITH ONE HAND, LIFTS R FOOT, 3 SECONDS	0☐	1☐	2☐	3☐	54.
*	55.	STD: HOLDING ON TO LARGE BENCH WITH ONE HAND, LIFTS L FOOT, 3 SECONDS	0☐	1☐	2☐	3☐	55.
*	56.	STD: MAINTAINS, ARMS FREE, 20 SECONDS	0☐	1☐	2☐	3☐	56.
*	57.	STD: LIFTS L FOOT, ARMS FREE, 10 SECONDS	0☐	1☐	2☐	3☐	57.
*	58.	STD: LIFTS R FOOT, ARMS FREE, 10 SECONDS	0☐	1☐	2☐	3☐	58.
*	59.	SIT ON SMALL BENCH: ATTAINS STD WITHOUT USING ARMS	0☐	1☐	2☐	3☐	59.
*	60.	HIGH KN: ATTAINS STD THROUGH HALF KN ON R KNEE, WITHOUT USING ARMS	0☐	1☐	2☐	3☐	60.
*	61.	HIGH KN: ATTAINS STD THROUGH HALF KN ON L KNEE, WITHOUT USING ARMS	0☐	1☐	2☐	3☐	61.
*	62.	STD: LOWERS TO SIT ON FLOOR WITH CONTROL, ARMS FREE	0☐	1☐	2☐	3☐	62.
*	63.	STD: ATTAINS SQUAT, ARMS FREE	0☐	1☐	2☐	3☐	63.
*	64.	STD: PICKS UP OBJECT FROM FLOOR, ARMS FREE, RETURNS TO STAND	0☐	1☐	2☐	3☐	64.

TOTAL DIMENSION D ☐

Item		E: WALKING, RUNNING & JUMPING	SCORE				NT
*	65.	STD, 2 HANDS ON LARGE BENCH: CRUISES 5 STEPS TO R	0☐	1☐	2☐	3☐	65.
*	66.	STD, 2 HANDS ON LARGE BENCH: CRUISES 5 STEPS TO L	0☐	1☐	2☐	3☐	66.
*	67.	STD, 2 HANDS HELD: WALKS FORWARD 10 STEPS	0☐	1☐	2☐	3☐	67.
*	68.	STD, 1 HAND HELD: WALKS FORWARD 10 STEPS	0☐	1☐	2☐	3☐	68.
*	69.	STD: WALKS FORWARD 10 STEPS	0☐	1☐	2☐	3☐	69.
*	70.	STD: WALKS FORWARD 10 STEPS, STOPS, TURNS 180°, RETURNS	0☐	1☐	2☐	3☐	70.
*	71.	STD: WALKS BACKWARD 10 STEPS	0☐	1☐	2☐	3☐	71.
*	72.	STD: WALKS FORWARD 10 STEPS, CARRYING A LARGE OBJECT WITH 2 HANDS	0☐	1☐	2☐	3☐	72.
*	73.	STD: WALKS FORWARD 10 CONSECUTIVE STEPS BETWEEN PARALLEL LINES 20cm (8")APART	0☐	1☐	2☐	3☐	73.
*	74.	STD: WALKS FORWARD 10 CONSECUTIVE STEPS ON A STRAIGHT LINE 2cm (3/4") WIDE	0☐	1☐	2☐	3☐	74.
*	75.	STD: STEPS OVER STICK AT KNEE LEVEL, R FOOT LEADING	0☐	1☐	2☐	3☐	75.
*	76.	STD: STEPS OVER STICK AT KNEE LEVEL, L FOOT LEADING	0☐	1☐	2☐	3☐	76.
*	77.	STD: RUNS 4.5m (15'), STOPS & RETURNS	0☐	1☐	2☐	3☐	77.
*	78.	STD: KICKS BALL WITH R FOOT	0☐	1☐	2☐	3☐	78.
*	79.	STD: KICKS BALL WITH L FOOT	0☐	1☐	2☐	3☐	79.
*	80.	STD: JUMPS 30cm (12") HIGH, BOTH FEET SIMULTANEOUSLY	0☐	1☐	2☐	3☐	80.
*	81.	STD: JUMPS FORWARD 30 cm (12"), BOTH FEET SIMULTANEOUSLY	0☐	1☐	2☐	3☐	81.
*	82.	STD ON R FOOT: HOPS ON R FOOT 10 TIMES WITHIN A 60cm (24") CIRCLE	0☐	1☐	2☐	3☐	82.
*	83.	STD ON L FOOT: HOPS ON L FOOT 10 TIMES WITHIN A 60cm (24") CIRCLE	0☐	1☐	2☐	3☐	83.
*	84.	STD, HOLDING 1 RAIL: WALKS UP 4 STEPS, HOLDING 1 RAIL, ALTERNATING FEET	0☐	1☐	2☐	3☐	84.
*	85.	STD, HOLDING 1 RAIL: WALKS DOWN 4 STEPS, HOLDING 1 RAIL, ALTERNATING FEET	0☐	1☐	2☐	3☐	85.
*	86.	STD: WALKS UP 4 STEPS, ALTERNATING FEET	0☐	1☐	2☐	3☐	86.
*	87.	STD: WALKS DOWN 4 STEPS, ALTERNATING FEET	0☐	1☐	2☐	3☐	87.
*	88.	STD ON 15cm (6") STEP: JUMPS OFF, BOTH FEET SIMULTANEOUSLY	0☐	1☐	2☐	3☐	88.

TOTAL DIMENSION E ☐

Was this assessment indicative of this child's "regular" performance? YES ☐ NO ☐

COMMENTS:

GMFM-88 SUMMARY SCORE

DIMENSION	CALCULATION OF DIMENSION % SCORES				GOAL AREA (indicated with ✓ check)
A. Lying & Rolling	Total Dimension A / 51	= ____ / 51	× 100 =	________ %	A. ☐
B. Sitting	Total Dimension B / 60	= ____ / 60	× 100 =	________ %	B. ☐
C. Crawling & Kneeling	Total Dimension C / 42	= ____ / 42	× 100 =	________ %	C. ☐
D. Standing	Total Dimension D / 39	= ____ / 39	× 100 =	________ %	D. ☐
E. Walking, Running & Jumping	Total Dimension E / 72	= ____ / 72	× 100 =	________ %	E. ☐

TOTAL SCORE = %A + %B + %C + %D + %E / Total # of Dimensions

= ________________ / 5 = ________ = ________ %

GOAL TOTAL SCORE = Sum of %scores for each dimension identified as a goal area / # of Goal areas

= ________________ = ________ %

GMFM-66 Gross Motor Ability Estimator Score [1]

GMFM-66 Score = ________________ ___________ to __________
95% Confidence Intervals

previous GMFM-66 Score = ________________ ___________ to __________
95% Confidence Intervals

change in GMFM-66 = ________________

[1] from the Gross Motor Ability Estimator (GMAE-2) Software

TESTING WITH AIDS/ORTHOSES USING THE GMFM-88

Indicate below with a check (4) which aid/orthosis was used and what dimension it was first applied. (There may be more than one).

AID	Dimension		Orthosis	Dimension	
Rollator/pusher	☐	______	Hip Control..	☐	______
Walker..	☐	______	Knee Control.......................................	☐	______
H Frame crutches	☐	______	Ankle-foot Control	☐	______
Crutches	☐	______	Foot Control	☐	______
Quad Cane	☐	______	Shoes..	☐	______
Cane	☐	______	None	☐	______
None	☐	______	Other ______ (please specify)	☐	______
Other ______ (please specify)	☐	______			

GMFM-88 SUMMARY SCORE USING AIDS/ORTHOSES

DIMENSION	CALCULATION OF DIMENSION % SCORES				GOAL AREA (indicated with ✓ check)
F. Lying & Rolling	Total Dimension A / 51	= ______ / 51	× 100 =	______ %	A. ☐
G. Sitting	Total Dimension B / 60	= ______ / 60	× 100 =	______ %	B. ☐
H. Crawling & Kneeling	Total Dimension C / 42	= ______ / 42	× 100 =	______ %	C. ☐
I. Standing	Total Dimension D / 39	= ______ / 39	× 100 =	______ %	D. ☐
J. Walking, Running & Jumping	Total Dimension E / 72	= ______ / 72	× 100 =	______ %	E. ☐

TOTAL SCORE = $\frac{\%A + \%B + \%C + \%D + \%E}{\text{Total \# of Dimensions}}$

= $\frac{\_\_\_\_\_\_}{5}$ = ______ = ______ %

GOAL TOTAL SCORE = $\frac{\text{Sum of \%scores for each dimension identified as a goal area}}{\text{\# of Goal areas}}$

= ______ = ______ %

iv. Edinburgh Visual Gait Score

The Edinburgh Visual Gait Score (EVGS) is a visual gait assessment scale and was created and validated for use in children with CP by Read, Hazlewood, Hillman, Prescott, and Robb (2003). Their intent was for this tool to be a valid and reliable visual gait scoring system that would be practical for clinical use. This was confirmed by a feasibility study by Gupta & Raja (2012), who reported the EVGS can be used as an outcome measure to assess effectiveness of an intervention.

Please see below for the original EVGS assessment sheet used. MCID value of 2.4 is found for the EVGS; representing the improvement in gait score that is likely to reflect a clinical improvement in function (Robinson, Clement, Herman, & Gaston, 2017).

Edinburgh Visual Gait Score for Use in Cerebral Palsy

Explanatory Notes

The P&O Clinical Movement DATA software can assist you in calculating the Edinburgh Visual Gait Score (EVGS). Use the on-screen measurement and drawing tools to help you to evaluate each item of the score and enter the results in the EVGS pane on the left hand side of the screen. The software will then generate a EVGS report, with illustrative images from the video if desired, in PDF format.

Foot

1. Initial Contact in Stance

The heel normally contacts first. The toe describes that portion of the foot distal to the metatarso-phalangeal joints. Simultaneous contact with the heel and toe comprises flatfoot contact.

Observation	Score
Heel contact	0
Flatfoot contact	1
Toe contact	2

2. Heel Lift in Stance

If there is no heel contact during stance, there can be no heel lift (i.e., "No heel contact"). Heel lift normally occurs between opposite foot level and opposite foot contact ("Normal"). "Early" heel lift indicates that heel lift precedes the opposite foot being level with the stance foot. "Delayed" heel lift is present if heel lift occurs with or after opposite foot contact. "No forefoot contact" describes the rare occasion of a calcaneous foot when the forefoot does not contact during stance.

Observation	Score
No forefoot contact	2
Delayed	1
Normal	0
Early	1
No heel contact	2

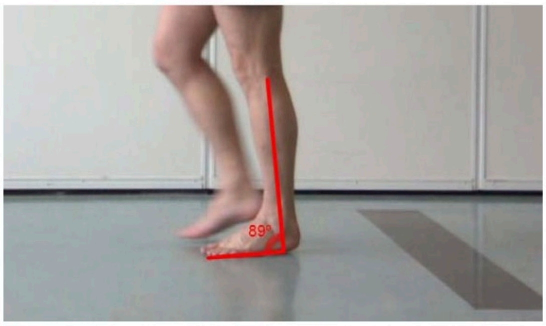
3. Maximum Ankle Dorsiflexion in Stance

There is normal forward progression of the tibial over the planted hindfoot from slight plantarflexion at initial contact to dorsiflexion at terminal stance.

Describe the maximum angle of dorsiflexion between hindfoot and shaft of the tibia during stance.

In pathological gait, lack of heel contact may be caused by either excessive plantarflexion of the foot or excessive knee flexion. The tibial-hindfoot angle is therefore analysed irrespective of the position of the foot on the floor.

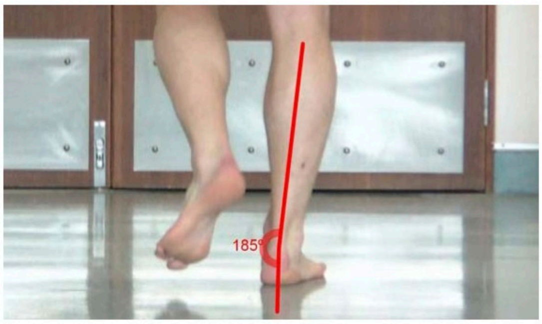
Observation	Score
Excessive dorsiflxn (>40° df)	2
Increased dorsiflxn (26°- 40° df)	1
Normal dorsiflxn (5°- 25° df)	0
Reduced dorsiflxn (10° pl - 4° df)	1
Marked plantarflxn (>10° pl)	2



4. Hind-foot Varus/Valgus in Stance

In the coronal plane, the normal hind-foot is in neutral or very slight valgus.

Observation	Score
Severe valgus (more than 15° valgus)	2
Mod valgus (6° to 15° valgus)	1
Neutral/slight valgus (0° to 5° valgus)	0
Mild varus (1° to 10° varus)	1
Severe varus (more than 10° varus)	2



5. Foot Rotation in Stance

The normal foot is slightly externally rotated relative to the Knee Progression Angle (KPA, i.e., the direction in which the knee points during gait).

Observation	Score
Marked ext >KPA (by >40°)	2
Mod ext >KPA (by 21°- 40°)	1
Sl more ext than KPA (by 0°- 20° extn)	0
Mod int >KPA (by 1°- 25°)	1
Marked int >KPA (by >25°)	2

6. Clearance in Swing

The whole foot including the toe should clear the foot and not make contact during swing phase.

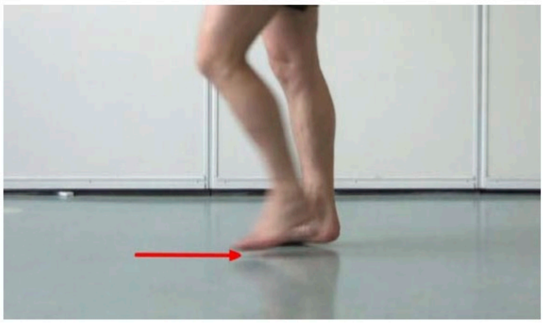
"None" should be recorded if there is continuous contact between some part of the foot and the floor throughout swing phase.

"Reduced" indicates that there is a shortened but definite period of clearance during some part of the swing phase between the whole foot and the floor.

"Full" or normal clearance is when the foot does not touch at all in swing; however, normal clearance is a very small amount.

"High steps" describes excessive lifting of the foot from the floor. When there is reduced clearance followed by high stepping, circle both, giving a score of 2 for this combination of features.

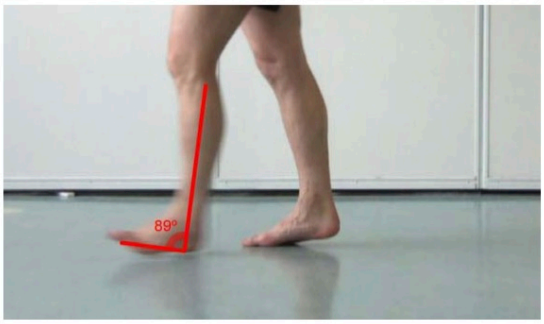
Observation	**Score**
High Steps	1
Full	0
Reduced	1
None	2



7. Maximum Ankle Dorsiflexion in Swing

The ankle is normally approximately neutral in swing, but very slight plantarflexion (5°) is acceptable.

Observation	**Score**
Excessive dorsiflxn (>30° df)	2
Increased dorsiflxn (16°-30° df)	1
Normal dorsiflxn (15° df- 5° pl)	0
Mod plantarflxn (6°- 20° pl)	1
Marked plantarflxn (>20° pl)	2



Knee

8. Knee Progression Angle in Mid-Stance

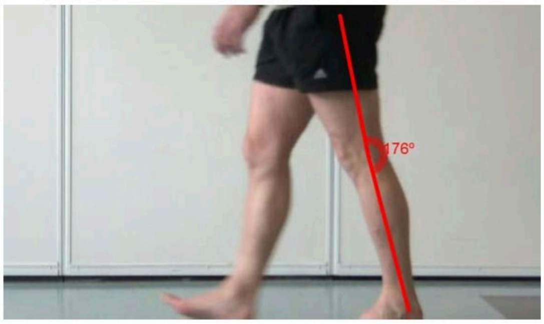
The knee normally points forward during gait. Record the position in which the knee appears to point during most of the stance phase. When either internal or external rotation is present but the whole knee cap is visible, score 1. When rotation is present to such an extent that the knee cap is partially out of view (external or internal, part cap visible), score 2.

Observation	Score
External, part knee cap visible	2
External, all knee cap visible	1
Neutral, knee cap midline	0
Internal, all knee cap visible	1
Internal, part knee cap visible	2

9. Peak Extension Stance

The knee approaches full extension in terminal stance. In pathological gait, the knee may remain more flexed throughout stance. Alternatively, hypertension can occur as femoral progression proceeds over an arrested tibia.

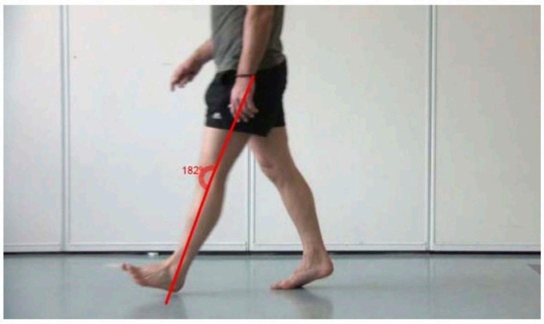
Observation	Score
Severe flexn (>25°)	2
Mod flexn (16°- 25°)	1
Normal (0°- 15° flexn)	0
Mod hyperextn (1°- 10°)	1
Severe hyperextn (<10°)	2



10. Terminal Swing Position

The knee is normally in slight flexion immediately before heel strike.

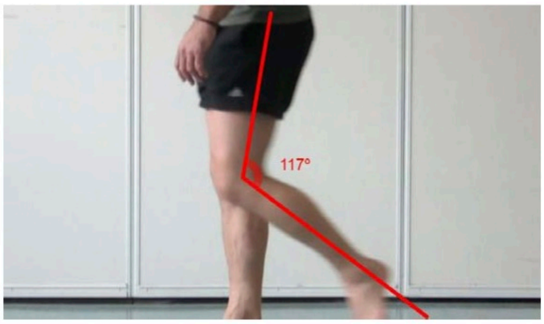
Observation	Score
Severe flexn (>30°)	2
Mod flexn (16°- 30°)	1
Normal (5°- 15° flxn)	0
Mod overextn (4° flx- 10° xtn)	1
Severe hyperextn (>10° xtn)	2



11.Peak Knee Flexion in Swing

The normal range is 50° to 70°.

Observation	Score
Severely increased (>85° flxn)	2
Mod increased (71°- 85° flxn)	1
Normal (50°- 70° flxn)	0
Mod reduced (35°- 49° flxn)	1
Severely reduced (<35° flxn)	2

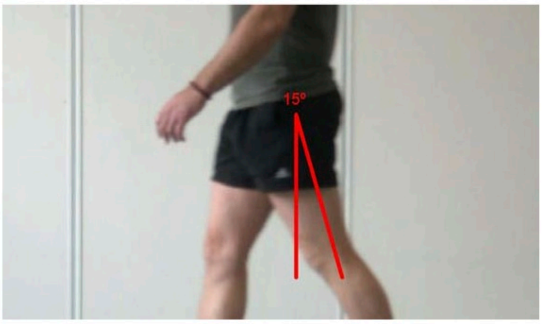


Hip

12.Peak Hip Extension in Stance

The hip normally extends in stance to between neutral and 20° of extension.

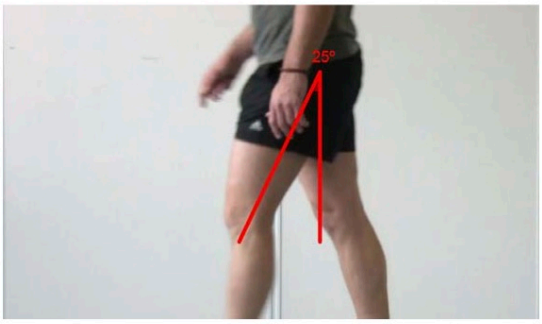
Observation	Score
Mod flexn (°1- 15° flxn)	2
Mod flexn (°1- 15° flxn)	1
Normal (0°- 20° extn)	0
Mod hyperextn (21°- 35° extn)	1
Marked hyperextn (>35°)	2



13.Peak Hip Flexion during Swing

Normal flexion is between 25° and 45°.

Observation	Score
Marked increase (>60° flxn)	2
Increased flexn (46°- 60° flxn)	1
Normal flexn (25°- 45° flxn)	0
Reduced flexn (10°- 24° flxn)	1
Severely reduced (<10° flxn)	2



Pelvis

14.Obliquity at Mid-Stance

The pelvis normally drops slightly on the opposite side during loading, becoming level by terminal stance. Estimate the position in mid stance. "Up" and "down" refer to the position of the ASIS on the stance side, relative to the opposite side ASIS.

Observation	Score
Marked down (>10°)	2
Mod down (1°- 10°)	1
Normal obliquity (0°- 5° up)	0
Mod up (6°- 15°)	1
Marked up (>15°)	2

15.Pelvic Rotation at Mid-Stance

In mid stance, the pelvis should be at approximately neutral rotation, between 5° backward rotation (retraction) of the stance leg, and 10° forward rotation (protraction).

Observation	Score
Marked retraction (>15°)	2
Mod retraction (6°- 15°)	1
Normal (5° retr- 10° pro)	0
Mod protraction (11°- 20°)	1
Severe protraction (>20°)	2

Trunk

16.Peak Sagittal Position in Stance

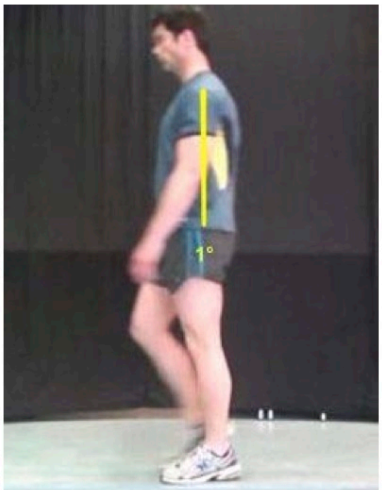
The trunk is erect during stance and swing phases. Suggested values here are:

0 = Vertical to 5° forward or backward

1 = more than 5° backwards or between 6° and 15° forward

2 = more than 15° forward

Observation	Score
Marked	2
Moderate	1
Normal	0
Reduced	1



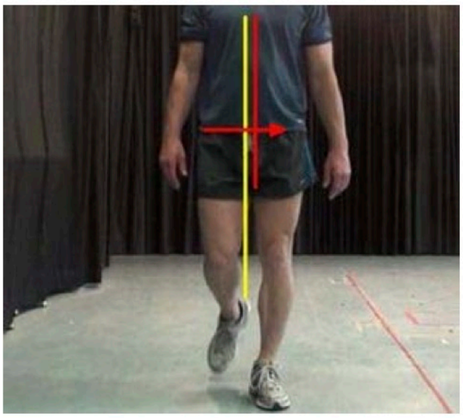
17.Maximum Lateral Shift

Normally the trunk displaces laterally approximately 25 mm during stance, towards the stance leg.

"Excessive" thoracic shift laterally or lateral flexion should be considered when recording observations.

"Reduced" describes those cases in which the trunk remains leaning over the swinging leg.

Observation	Score
Excessive	2
Normal	0
Reduced	1



v. Cerebral Palsy Quality of Life Questionnaire (CPQoL)

Please visit the following webpage for full selection of CPQoL.
https://www.ausacpdm.org.au/research/cpqol/

vi. Questionnaire (Q'AIRE)

A questionnaire (Q'AIRE) was designed and developed specifically to focus on AFOs and SMotOs (please see below for full Q'AIRE). Prior to administering the Q'AIRE for parents, the tool was reviewed by two senior research academics. Finke (2005) recommends this process as a means of increasing survey reliability. The purposes of the reviews were to (a) evaluate the effectiveness of the survey for capturing required data; and (b) obtain feedback regarding the investigator-designed questions. The feedback provided by reviewers were considered against the intended scope of the research questions in order to inform revisions to the Q'AIRE for parents tool and as such would maximise the external validity of the information captured and the results generated by the questionnaires; a method advised by Fink (2005).

Please see following for full Q'AIRE;

Project Number:	RO-1835
Project Title:	The effect of ankle –foot orthoses vs. sensomotoric orthoses on gait, gross motor skills and quality of life in children with cerebral palsy
Researcher Supervisors:	Professor Wayne Hing Professor of Physiotherapy, Faculty HSM, Bond University Assistant Professor Rob Orr Physiotherapy, Faculty HSM, Bond University
Principal Researcher:	Ms Clare MacFarlane (PhD Candidate), DPhty, B Sc (Sp. & Ex.). Physiotherapist, NAPA Centre
Co-Investigators:	David Wong Podiatrist, Better Health

Aims of the questionnaire

This questionnaire aims to determine the parent / caregiver perspective and feedback on differences in gait, gross motor skills and quality of life when their children with cerebral palsy are wearing AFO's vs. SMO's.

Children with cerebral palsy (CP) have many gait anomalies and gross motor deficits, which may affect quality of life. Most of the gait anomalies are addressed with the use of Ankle-Foot Orthoses (AFO's). An alternate treatment option has been the use of Sensomotoric Orthotics (SMotO's) to assist gait. SMotO's are a dynamic type of orthotic, designed to activate certain muscles via bumps that coincide with these muscles in the foot.

Parent Questionnaire;

Thank you for taking the time to answer the following questions. Your feedback and honest opinion will help future research and decision making for other families considering Sensomotoric Orthotics as a treatment option for their child.

Please answer the following questions by **bolding** your answer and filling out where appropriate. There are two (2) sections; Section 1 relates to Ankle- Foot Orthoses (AFOs), and Section 2 relates to Sensomotoric Orthotics (SMotO)

SECTION 1: AFOs

1. **At what age did your child begin to wear AFOS?**
    a. Age:

2. **What reasons were given for their prescription? (can answer more than one)**
    a. Unknown / none given
    b. Tightness in calves / ankles (for a stretch)
    c. Balance
    d. Walking ability
    e. Alignment
    f. Because they have cerebral palsy
    g. Other (please describe)?

3. **Were videos taken or footprints measured and/or remeasured at the time?**
    a. No
    b. Yes
        i. If yes, which type?

4. **Was the time process between AFO casting and taking them home with you?**
    a. One month
    b. Two months
    c. Three months
    d. Between 3-6 months
    e. Over 6 months

5. **What type of AFO was your child given?**
    a. Hinged AFO
    b. Solid AFO
    c. Dynamic AFO
    d. Posterior Leaf Spring
    e. Other

6. **Did you need or have subsequent appointments for fittings or adjustments once you had the AFOs?**
    a. Yes
    b. No

7. **How long has your child been wearing AFO's for now?**
    a. Less than one year
    b. Between one and two years

c. Between two and three years
d. Over three years
e. Doesn't wear them anymore
f. Other

8. Is your child comfortable wearing their AFOs?
a. Generally Comfortable
b. Comfortable
c. Generally Uncomfortable
d. Uncomfortable
e. Refuses to wear them
f. Other (please describe)

9. Does your child experience any pain wearing AFO?
a. Yes / no / sometimes

10. Does their discomfort affect or dictate if your child wears the AFO?
a. Yes
b. No

11. How long does your child wear AFOs for throughout the day?
a. Less than one hour
b. Between one and two hours
c. Two to three hours
d. Three to four hours
e. Four to six hours
f. Over six hours

12. When wearing the AFOs, have you noticed an improvement in their (can answer more than one);
a. Sitting on the floor
b. Crawling
c. Transitions into standing
d. Balance when standing still
e. Alignment in their legs
f. Walking
    i. distance
    ii. endurance
    iii. balance
    iv. speed
g. other? (please describe)

13. How do you feel AFO's benefit your child?

a. Alignment
b. Balance
c. Walking ability
d. Stretch
e. Other?

14. What do you DISLIKE about AFO's?

a. Bulk
b. Fitting into shoes
c. The way they look
d. Restriction in movement
e. Difficulty to put on
f. Pressure areas
g. Exclusion from activities or participation at school
h. Other (please describe)

15. Were you ever offered an alternative to AFOs?

a. No
b. Yes
    i. If yes, what were they?

SECTION 2: SMotOs

1. **How long has your child had the SMotOs?**
    a. Less than one year
    b. Between one and two years
    c. Between two and three years
    d. Over three years
    e. Doesn't wear them anymore
        i. If not, why?
    f. Other

2. **Does your child wear both AFOs and SMotOs?**
    a. No
    b. Yes
        i. If yes, what is the use time split between them?
            1. (AFO) 0 / 100 (SMotO)
            2. (AFO) 20 / 80 (SMotO)
            3. (AFO) 30 / 70 (SMotO)
            4. (AFO) 40 / 60 (SMotO)
            5. (AFO) 50 / 50 (SMotO)
            6. (SMotO) 0 / 100 (AFO)
            7. (SMotO) 20 / 80 (AFO)

8. (SMotO) 30 / 70 (AFO)
9. (SMotO) 40 / 60 (AFO)
10. Other (please define)?

3. **When wearing the SMotOs, have you noticed an improvement in their (can answer more than one);**
    a. Sitting on the floor
    b. Crawling
    c. Transitions into standing
    d. Balance when standing still
    e. Alignment in their legs
    f. Walking
        i. distance
        ii. endurance
        iii. balance
        iv. speed
    g. other? (please describe)

4. **Do you see a difference in your child's ability for inclusion when wearing the SMotO vs the AFO?**
    a. No
    b. Yes
        i. Therapy
        ii. Play with friends / siblings
        iii. Inclusion in activities at school
        iv. Other? (please describe)

5. **What do you DISLIKE about SMotO?**
    a. Discomfort
    b. Fitting in shoes
    c. Doesn't give a stretch
    d. Pressure areas
    e. Other (please describe)?

6. **Do you wish you had more orthotics options on offer in the public system?**
    a. No, I'm happy with what has been offered
    b. Yes, I would like to be shown alternate options for my child

Is there anything else you would like to comment on in regard to your experience with either AFOs or SMotOs?

e. Results of Q'AIRE (Study 7)

AFO Question	Answer Options	AFO Average or Collated Response
1. At what age did your child begin to wear AFOS?	N/A	Average = 2.5 years
2. What reasons were given for their prescription? (can answer more than one)	a. Unknown / none given b. Tightness in calves / ankles (for a stretch) c. Balance d. Walking ability e. Alignment f. Because they have cerebral palsy g. Other (please describe)?	a. = 0 b. = 6 (37.5%) c. = 5 (31.3%) d. = 6 (37.5%) e. = 7 (43.8%) f. = 6 (37.5%) g. = 2 (12.5%) To help reduce pointing of toes Reduce ankle rolling
3. Were videos taken or footprints measured and/or remeasured at the time?	a. No b. Yes i. If yes, which type?	a. = 16 (100%) b. = 0
4. What was the time process between AFO casting and taking them home with you?	a. One month b. Two months c. Three months d. Between 3-6 months e. Over 6 months	a. = 11 (68.8%) b. = 4 (25%) c. = 0 d. = 1 (6.3%) e. = 0
5. What type of AFO was your child given?	a. Hinged AFO b. Solid AFO c. Dynamic AFO d. Posterior Leaf Spring e. Other	a. = 2 (12.5%) b. = 14 (87.5%) c. =1 (6.3%) d. = 0 e. = 0
6. Did you need or have subsequent appointments for fittings or adjustments once you had the AFOs?	a. Yes b. No	a. = 12 (75%) b. = 4 (25%)
7. How long has your child been wearing AFO's for now?	a. Less than one year b. Between one and two years c. Between two and three years d. Over three years e. Doesn't wear them anymore f. Other	a. =1 (6.3%) b. =1 (6.3%) c. = 2 (12.5%) d. = 5 (31.3%) e. = 6 (37.5%) f. = 2 (12.5%) Hasn't worn them for approx. 3months Uses them in combo with SMotO
8. Is your child comfortable wearing their AFOs?	a. Comfortable b. Mix of comfortable and uncomfortable c. Uncomfortable d. Refuses to wear them e. Other (please describe)	a. = 5 (31.3%) b. = 7 (43.8%) c. = 2 (12.5%) d. = 3 (18.8%) e. = 1 (just got new ones from USA)
9. Does your child experience any pain wearing AFO?	a. Yes b. No c. Sometimes	a. = 3 (18.8%) b. = 5 (31.3%) c. = 8 (50%) At feet

		When getting too tight / small Toes Ankle / arch Top/back of calf where they rub against skin High tone and dystonia meant she could push out of them; her heel would move and then the hard plastic pushed everywhere it shouldn't Not sure where although cries when wearing AFOs
10. Does their discomfort affect or dictate if your child wears the AFO?	a. Yes b. No c. N/A	a. = 8 (50%) b. = 6 (37.5%) c. = 2 (12.5%)
11. How long does your child wear AFOs for throughout the day?	a. Less than one hour b. Between one and three hours c. Three to six hours d. Over six hours e. N/A = 1	a. = 2 (12.5%) b. = 2 (12.5%) c. = 6 (37.5%) d. = 5 (31.3%)
12. When wearing the AFOs, have you noticed an improvement in their (can answer more than one);	a. Sitting on the floor b. Crawling c. Transitions into standing d. Balance when standing still e. Alignment in their legs f. Walking i. distance ii. endurance iii. balance iv. speed g. other? (please describe)	a. = 0 b. = 0 c. = 2 (12.5%) d. = 8 (50%) e. = 8 (50%) f. = 7 (43.8%) i. = 3 ii. = 2 iii. = 3 iv. = 2 Straighter when strapped into standing frame or wheelchair Stopped feet from rolling slightly
13. How do you feel AFO's benefit your child?	a. Alignment b. Balance c. Walking ability d. Calf length/stretch e. Prevented surgery f. Other?	a. = 8 (50%) b. = 7 (43.8%) c. = 5 (31.3%) d. = 6 (37.5%) e. = 1 (6.3%) f. = 2 (12.5%) Reduced ankle rolling slightly so gave slightly better step Support to carer with transfers
14. What do you DISLIKE about AFO's?	a. Bulky b. Fitting into shoes c. The way they look d. Restriction in movement e. Difficulty to put on f. Pressure areas g. Exclusion from activities or participation at school h. Other (please describe)	a. = 9 (56.3%) b. = 10 (62.5%) c. = 4 (25%) d. = 15 (93.8%) e. = 6 (37.5%) f. = 8 (50%) g. = 3 (18.8%) h. = 3 (18.8%) Lack of sensory input from feet, lack of development of ankle and calf muscle

			Getting into the wheelchair is difficult, not being able to sit cross legged comfortably on floor Inability to take steps in any way, unable to sit on floor comfortably
15.	Were you ever offered an alternative to AFOs?	a. No b. Yes If yes, what were they?	a. = 16 (100%) b. = 0 (Parents reported being offered SMotO and Piedro boots via therapy clinic, not through public system where they are prescribed AFOs)

f. Approval Letters

BOND UNIVERSITY
BRINGING AMBITION TO LIFE

HUMAN RESEARCH ETHICS COMMITTEE
Bond University
Gold Coast, Queensland 4229
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Fax: +61 7 5595 1120
(from overseas)
Email: buhrec@bond.edu.au
ABN 88 010 694 121
CRICOS CODE 00017B

12 October 2012

Dr. Wayne Hing
Faculty of Health Sciences & Medicine
Bond University

Dear Wayne

Protocol No: **RO 1539**
Project Title: **Inter- and Intra-tester reliablility of ALM**

I am pleased to confirm that your project was reviewed under the Full review procedure of Bond University's Human Research Ethics Committee and you have been granted approval to proceed.

As a reminder, BUHREC's role is to monitor research projects until completion. The Committee requires, as a condition of approval, that all investigations be carried out in accordance with the National Health and Medical Research Council's (NHMRC) National Statement on Ethical Conduct in Research Involving Humans and Supplementary Notes. Specifically, approval is dependent upon your compliance, as the researcher, with the requirements set out in the National Statement as well as the research protocol and listed in the Declaration which you have signed.

Please be aware that the approval is given subject to the protocol of the study being undertaken as described in your application with amendments, where appropriate. As you may be aware the Ethics Committee is required to annually report on the progress of research it has approved. We would greatly appreciate if you could advise us when you have completed data collection and when the study is completed

Should you have any queries or experience any problems, please contact early in your research project: Telephone: (07) 559 53554, Facsimile: (07) 559 51120, Email: buhrec@bond.edu.au.

We wish you well with your research project.

Yours sincerely

Dr Mark Bahr
Chair

www.bond.edu.au

BOND UNIVERSITY
BRINGING AMBITION TO LIFE

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ABN 88 010 694 121
CRICOS CODE 00017B

18 August 2014

Wayne Hing, Claire MacFarlane, Rob Orr and David Wong
Faculty of Health Science and Medicine
Bond University

Dear Wayne, Claire, Rob and David

Protocol No: **RO 1835**
Project Title: **The effect of AFO's vs. SMO's on gait, gross motor skills and quality of life in children with Cerebral Palsy**

I am pleased to confirm that your project was reviewed under the Full review procedure of Bond University's Human Research Ethics Committee and you have been granted approval to proceed.

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Should you have any queries or experience any problems, please contact early in your research project: Telephone: (07) 559 53554, Facsimile: (07) 559 51120, Email: buhrec@bond.edu.au.

We wish you well with your research project.

Yours sincerely

M Bahr

Dr Mark Bahr
Chair

www.bond.edu.au